THE UNIVERSITY *of* EDINBURGH

HEALTH-RELATED DECISION-MAKING IN ITS PERSONAL, SOCIAL AND HEALTH SERVICE CONTEXTS:

A critical review of relevant findings from seven publications and consideration of their contribution to understandings of decision-making and the wider field of applied health services research

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DECLARATION

I declare that I have composed this thesis (critical review) and made a substantial contribution to all the published papers included in the accompanying portfolio. The nature of that contribution is clearly detailed within the critical review. I further declare that this work has not been submitted for any other degree or professional qualification.

Ruth I. Hart

ABSTRACT

Health-related decision-making, in particular patients' involvement in decision-making about their treatment and care, has been an important and enduring concern for many practitioners and researchers working in applied health services research and allied fields. This is evidenced by the substantial (and still growing) body of work on 'shared decision-making' (SDM). With the aim of advancing understandings of decision-making, and the associated literature, this critical review seeks to situate, present, draw together, and critically consider, relevant findings from work (seven papers) I have first-authored. These papers arose from three applied (qualitative) health services research studies which directly or indirectly explored the experiences of different groups of patients confronted with decisions about their treatment and/or care.

I begin my review with a short overview of relevant theoretical and empirical work pre-dating and informing my own research studies and publications, noting some emergent critiques, and highlighting where important gaps in evidence and understanding were said, at the time, to remain. Then, shifting focus to my own work, I introduce the three studies from which the submitted publications arose, detailing their backgrounds, aims, methods and my involvement in each.

Next, I reflect on the findings of my submitted papers, noting how individually and collectively they indicate the importance of the context(s) in which health-related decisions are made. Using techniques of qualitative synthesis to identify a series of descriptive and analytic themes, I develop – and evidence – the proposition that health-related decision-making happens in, and is shaped by, its personal, social and health service contexts. This includes detailing the various ways in which different features of context may influence patients' decision-making.

I then consider, critically, how my findings fit with the wider literature. I proceed to argue that, in attending to, and highlighting, the role of context, my papers, synthesis and review provide insights that complement and extend the historic emphasis in SDM scholarship on what goes on within clinical encounters. Reflecting on the focus of more recent SDM literature (publications contemporaneous with or subsequent to work leading to the submitted papers) I note where other authors have similarly gone on to assert the importance of taking a more context-cognisant approach to

understanding health-related decision-making. I also consider how other literatures (such as the classic literature of medical sociology and more recent work in psychology) support and might usefully inform this and future thinking about health-related decision-making.

Moving the review towards a close, I offer an assessment of the strengths and limitations of my published work and, moreover, of my synthesis and review. I finish by reflecting upon the methodological and other learning I have accrued over the course of undertaking the contributing studies, including preparing the submitted publications, and from the process of producing this critical review.

LAY SUMMARY

How patients and health professionals make decisions about patients' treatment and care is a topic that interests many researchers. Numerous teams have written about the idea and realities of 'shared decision-making' (SDM) – where patients and professionals make these sorts of decisions in partnership. With a view to extending what is known about health-related decision-making (shared or otherwise) I have reviewed and reflected upon relevant work I personally undertook. That work comprised seven published papers linked to three research projects, each of which explored some aspect(s) of patients' experiences of health-related decision-making.

I begin my report on that review process by summarising important work that others had done ahead of my own, and which provided a foundation for me to build upon. I draw attention to some criticisms that had been made of that early work (for example, gaps and oversights that had been identified). I then write about my own work, outlining the three research projects which my published papers were linked to and explaining the role I played in each. Next, I discuss relevant findings from my published papers, and identify some prominent themes. I highlight how these point to the importance of the context(s) in which health-related decisions are made, and explain how my research suggests patients' decisions may be influenced by features of (a) their personal context, (b) their social context, and (c) the wider health service context in which they receive care.

Next, I consider how my findings 'fit' with those of other researchers. I suggest that in highlighting the important role of context, my findings complement and extend the historic focus on consultations (and the conversations between professionals and patients which take place within these). I note that in recent years some other researchers have similarly argued that we need to look more closely at wider influences on decision-making, including different types of context. I draw attention to work in other fields (in particular, medical sociology and psychology) that might help us understand different features of context and their influences better.

Finally, I wrap up my report by considering the strengths and weaknesses of my work (the original research, my published papers, and this review). I offer some ideas as to how research and practice might be different in the future. In addition, I

reflect on what I have learnt from undertaking the contributing work, and from the process of preparing and writing this review.

ACKNOWLEDGEMENTS

Many people – other social scientists, health professionals, and patients – enabled and/or supported the three studies and seven papers with which this review is concerned. I continue to be grateful for their contributions, which are acknowledged in the relevant paper(s). The support of two people has been particularly significant in the writing of this review: firstly – for assertive encouragement to complete the task, critical insights, and scholarly direction – Prof. Julia Lawton, with whom I have worked since late 2017; and, secondly, my partner, Tony Palmer – for patience, coffee, and a reliable supply of homemade cake.

PUBLICATIONS INCLUDED IN THE PORTFOLIO (and my contribution to each)

My portfolio comprises seven papers published 2015-2021, relating to research conducted between 2012 and 2019. These papers are reproduced in Appendix 3, with appropriate permissions. I am the first author of each though they are all – as is commonplace in applied health services research – co-authored publications. I detail my own contribution to each paper below. I discuss my wider role in the research projects from which the submitted papers arose in the following, introductory chapter.

1. Hart RI, Foster HE, McDonagh JE, Thompson B, Kay L, Myers A et al. Young people's decisions about biologic therapies: who influences them and how? *Rheumatology.* 2015; 54(7): 124-1301. https://doi.org/10.1093/rheumatology/keu523

I am this paper's first and corresponding author. I collected all the data it drew upon, led on the analysis, and conceived and drafted the manuscript.

2. Hart RI, McDonagh JE, Thompson B, Foster HE, Kay L, Myers A et al. Being as Normal as Possible: How Young People Ages 16–25 Years Evaluate the Risks and Benefits of Treatment for Inflammatory Arthritis. *Arthritis Care & Research*. 2016; 68(9): 1288-94. https://doi.org/10.1002/acr.22832

I am this paper's first and corresponding author. I collected all the data it drew upon, led on the analysis, and conceived and drafted the manuscript.

3. Hart RI, Ng WF, Newton JL, Hackett KL, Lee RP, Thompson B. What impact does written information about fatigue have on patients with auto-immune rheumatic diseases? Findings from a qualitative study. *Musculoskeletal Care*. 2017; 15(3): 230-237. http://dx.doi.org/10.1002/msc.1164

I am this paper's first and corresponding author. I collected all the data it drew upon, led on the analysis, and conceived and drafted the manuscript.

4. Hart RI, Hallowell N, Harden J, Jesudason AB, Lawton J. Clinician-researchers and custodians of scarce resources: A qualitative study of health professionals' views on barriers to teenagers and young adults' involvement in cancer trials. *Trials.* 2020; 21: 67. https://doi.org/10.1186/s13063-019-3942-y

I am this paper's first and corresponding author. I collected all the data it drew upon, led on the analysis, and conceived and drafted the manuscript.

5. Hart RI, Cameron DA, Cowie FJ, Harden J, Heaney NB, Rankin D et al. The challenges of making informed decisions about treatment and trial participation following a cancer diagnosis: a qualitative study involving adolescents and young adults with cancer and their caregivers. *BMC Health Services Research.* 2020; 20: 25. https://doi.org/10.1186/s12913-019-4851-1

I am the first author for this paper. I collected all the data it drew upon, and led on the analysis. I assisted with the initial drafting of the manuscript, and then revised that draft, quite substantially, in light of editor and reviewer comments, to produce the final, accepted version.

6. Hart RI, Cowie FJ, Jesudason AB, Lawton J. Adolescents and young adults' (AYA) views on their cancer knowledge prior to diagnosis: Findings from a qualitative study involving AYA receiving cancer care. *Health Expectations*. 2021; 24(2); 307-316. https://doi.org/10.1111/hex.13170

I am this paper's first and corresponding author. I collected all the data it drew upon, led on the analysis, and conceived and drafted the manuscript.

7. Hart RI, Boyle D, Cameron DA, Cowie FJ, Hayward L, Heaney NB et al. Strategies for improving access to clinical trials by teenagers and young adults with cancer: A qualitative study of health professionals' views. *European Journal of Cancer Care*. 2021; 30(3): e13408. https://doi.org/10.1111/ecc.13408

I am this paper's first and corresponding author. I collected all the data it drew upon, led on the analysis, and conceived and drafted the manuscript.

Other peer-reviewed publications, not included in the portfolio

The following papers were not included in my portfolio for one or more of the following reasons: their timing (i.e., recency of publication); relevance to health-related decision-making; and/or the nature and scale of my contribution.

First-authored publications

Hart RI, Kimbell B, Rankin D, Allen JM, Boughton CK, Campbell F et al. Parents' experiences of using remote monitoring technology to manage type 1 diabetes in very young children during a clinical trial: qualitative study. *Diabetic Medicine*. 2022; 39(7): e14828. https://doi.org/10.1111/dme.14828

Co-authored publications

Kimbell B, Rankin D, Hart RI, Allen JM, Boughton CK, Campbell F et al. Parents' views about healthcare professionals having real-time remote access to their young child's diabetes data: qualitative study. *Pediatric Diabetes.* 2022; (Online ahead of print). https://doi.org/10.1111/pedi.13363

Kimbell B, Rankin D, Hart RI, Allen JM, Boughton CK, Campbell F et al. Parents' experiences of using a hybrid closed-loop system (CamAPS FX) to care for a very young child with type 1 diabetes: qualitative study. *Diabetes Research and Clinical Practice.* 2022; 187. https://doi.org/10.1016/j.diabres.2022.109877

Norman JE, Lawton J, Stock SJ, Siassakos D, Norrie J, Hallowell N et al. Feasibility and design of a trial regarding the optimal mode of delivery for preterm birth: the CASSAVA multiple methods study. *Health Technology Assessment.* 2021; 25(61). https://doi.org/10.3310/hta25610

Lawton J, Hart RI, Kimbell B, Allen JM, Besser REJ, Boughton C et al. Data sharing whilst using a closed-loop system: Qualitative study of adolescents' and parents' experiences and views. *Diabetes Technology & Therapeutics.* 2021; 23(7): 500-507. https://doi.org/10.1089/dia.2020.0637

Howell DA, Hart RI, Smith A, Macleod U, Patmore R, Roman E. Unpacking pathways to lymphoma and myeloma diagnosis: Do experiences align with the Model of Pathways to Treatment? Findings from a UK qualitative study with patients and relatives. *BMJ Open.* 2020; 10(2): e034244. https://doi.org/10.1136/bmjopen-2019-034244

Howell DA, Hart RI, Smith AG, Macleod U, Patmore R, Roman E. Disease-related factors affecting timely lymphoma diagnosis: a qualitative study exploring patient experiences. *British Journal of General Practice*. 2019; 69(679): e134-e145. https://doi.org/10.3399/bjgp19X701009

Howell DA, Hart RI, Smith AG, Macleod U, Patmore R, Cook G et al. Myeloma: Patient accounts of their pathways to diagnosis. *PLOS ONE*. 2018; 13(4): e0194788. https://doi.org/10.1371/journal.pone.0194788

Hanson H, Hart RI, Thompson B, McDonagh JE, Tattersall R, Jordan A et al. Experiences of employment amongst young people with juvenile idiopathic arthritis: a qualitative study. *Disability and Rehabilitation*. 2018; 40(16): 1921-1928. https://doi.org/10.1080/09638288.2017.1323018

Lee RP, Hart RI, Watson RM, Rapley T. Qualitative synthesis in practice: some pragmatics of meta-ethnography. *Qualitative Research.* 2015; 15(3): 334-350. https://doi.org/10.1177/1468794114524221

CONTENTS

PREFACE

I begin with a brief account of how I came to undertake this review and the circumstances under which I completed it. My research career began 20 years ago when I joined a small private sector company providing policy-oriented research and evaluation services to the public and third sector. In 2008, I moved to a larger, third-sector organisation specialising in research and evaluation relating to education and children's services. This provided a stepping-stone to my first academic research post, applied health services research project, and experience of studies where (in addition to reports to funders) outputs were expected to include peer-reviewed publications. I have since worked as a research associate/fellow in three academic institutions: Newcastle University (2011-2015); the University of York (2015-2017); and the University of Edinburgh (2017-present).

As my time in, and commitment to, the field of applied (qualitative) health services research grew, I became increasingly keen, for pragmatic reasons, to secure recognition for my professional experience. A PhD by Research Publications offered both a route to accreditation and an opportunity to review and build upon prior work. In consultation with my line manager, Prof. Julia Lawton, I assessed whether I might compile selected work into a coherent portfolio, and undertake a sufficiently sophisticated cross-cutting analysis to warrant recognition as doctoral-level work. I subsequently registered for a PhD (by Research Publications), and with the support of my supervisors (Prof. Julia Lawton and Dr. Helen Eborall) commenced work on this review.

Notwithstanding the excellent intellectual, practical and emotional support provided by my advisors, the work has presented some challenges. The PhD by Research Publications route is an interesting one, but relatively unusual in the UK (Peacock, 2017). Comparable precedents are few and hard to locate, and University of Edinburgh guidelines are economical. The second challenge was one of context. I completed this work during the Covid-19 pandemic, whilst mostly working from home, personally experiencing a period of poor health, and juggling care responsibilities for elderly parents. It has at times been a welcome distraction, but at others felt quite a burden. I feel pride in what I have achieved in the circumstances.

BACKGROUND AND INTRODUCTION

Health-related decision-making, and in particular patients'[1] involvement in decision-making about their treatment and care, has been an important and enduring concern for many working in applied health services research and allied fields. This is borne out by the substantial – and still growing – body of work on 'shared decision-making' or 'SDM'. Much of my own work over the last decade has directly or indirectly explored health-related decision-making, from the perspectives of patients and others. In this critical review I seek to situate, present, draw together and critically consider relevant findings from that work, with the aim of advancing understandings of, and the literature on, health-related decision-making. This review is intended to be read in conjunction with the (seven) papers in my portfolio (see pp7-8 and Appendix 3); these arose from three separate applied (qualitative) health services research studies.

In this introductory chapter, I set the scene for my work, offering a brief overview of relevant theoretical and empirical literature predating and informing my own research and publications. I note some emergent critiques, and areas where important gaps in evidence and understanding were said, at the time, to exist. I then shift my focus to the original research underpinning my publications, introducing the three studies and detailing my involvement in each. In subsequent chapters I draw out and synthesise findings from my publications of salience to the topic of health-related decision-making, and consider how these complement and challenge the wider literature, including more recent SDM scholarship (i.e., publications contemporaneous with or subsequent to my own work) and work emerging from other fields.

Prior theoretical and empirical work informing my own research

Prior to my own research and writings, a large and diverse body of researchers (clinicians and academics) had undertaken work on health-related decision-making and the respective roles of clinicians and patients in this. Occupying a particularly

[1] I note that, though widely used in relevant literatures, the term 'patient' has come to be regarded in some quarters as problematic. I recognise there are arguments for using alternative terms, e.g., 'person' or 'research participant'. However, where writings/research also deal with the experiences and perspectives of other people and research participants (e.g., family members) these alternative terms need careful qualification to avoid ambiguity and confusion. I have reflected at length and, for reasons of specificity and economy, chosen to use the term 'patient' in this document where it facilitates understanding and readability.

important place in the growing corpus of relevant work was literature on, or speaking to, 'SDM'. Whilst the term SDM was coined in the 1980s (Katz, 1984), usage increased exponentially in the late 1990s, gaining momentum from diverse constituencies (e.g., doctors, patients, ethicists and researchers). Drivers included: evidence of variation in treatment practices, in response to similar problems; legislation affirming patients' rights; the consumer movement; and (feminist) challenges to medical authority (Charles et al., 1997 and 1999).

SDM came to be understood as the principle and practice of professionals and patients making (health-related) decisions collaboratively, i.e., in partnership. Though different models of SDM have since been proposed, the most widely cited remains that of Charles et al. (1997; 1999). This has three core features: bi-directional information exchange (clinicians sharing technical knowledge, and patients articulating their values and preferences); participation in deliberation by patients and clinicians; and consensus on the decision finally taken (Montori et al., 2006). Prior to my own research, the UK government had explicitly advocated SDM (Department of Health, 2010); many other administrations took a similar stance. Moreover, it had come to be largely accepted, in the literature, that in *most* scenarios, the ideal was for decisions to be shared, though SDM was surmised as being of particular relevance and importance in certain situations. These included where: there is clinical equipoise; the distribution of harms and benefits means the best choice is a matter of values and preferences; and/or implementation requires patients' commitment to action (Whitney, 2004; Elwyn et al., 2010).

Backtracking, however, there seems initially to have been more consensus about what SDM was *not* (traditional, 'paternalistic' decision-making, in which clinicians decided and patients acceded) than as to what it actually was. Indeed, Charles et al. (1997) explicitly cautioned that, though by then a widely used term, it was far from clear what SDM really meant and involved. Ensuing work sought to address that concern, by characterising and classifying SDM and other, adjacent forms of decision-making (e.g., informed decision-making) and their suitability for different scenarios (Gafni et al., 1998; Whitney, 2004). That work had impact, but new criticisms soon began to emerge. Gabe et al. (2004) highlighted the need for more empirical work to understand fully the practices involved; Montori et al. (2006) drew attention to differences in decision-making in acute and chronic care; and Edwards &

Elwyn (2006) argued for more 'patient-centred' research. Patient-centred(ness) is a term/concept predating but closely aligned with SDM; the two literatures are distinct, but overlapping, and in many respects have developed in parallel (see Box 1).

Patient-centred care: an allied area of research

As with SDM, the patient-centred literature and research focussed initially on physician-patient interactions within clinical encounters (in particular primary care consultations). Contrasted with traditional, 'disease-centred' approaches, patient-centred care involved developing – and responding to – an understanding of patients as people, and, moreover, 'see(ing) the illness through (their) eyes' (Levenstein et al., 1986: 26). Like the SDM literature, early writings focussed substantially on defining and measuring patient-centredness, in particular through observation and quantification of physician and patient behaviours (see, for example, Brown et al., 1986). Consistent with the concept, however, there was a relatively rapid shift towards canvassing patients' views – both on what patient-centredness meant and on other topics of research. This manifested in the increasing research attention given to 'softer' outcomes (such as satisfaction with care, and quality of life) which the patient perspective was essential to understanding (Laine & Davidoff, 1996). Whilst early studies largely tried to capture patients' views via quantitative methods and instruments (e.g., Stewart et al., 2000), it was soon recognised that important aspects of patient-centred care, and the patient experience more generally, might be more accessible via qualitative methods (Mead & Bower, 2000; Stewart, 2001).

Box 1

Moving forward, these calls for more empirical, diverse, and patient-centred research seem to have had an effect, with subsequent years seeing researchers publishing findings from empirical work, some qualitative, conducted in a multipliclity of clinical settings. These settings included general practice, psychiatry, obstetrics, and rheumatology (Saba et al. (2006); Seale et al. (2006); Emmett et al. (2006); Schildmann et al. (2008)). Research conducted around that time often involved close attention to: clinical communication processes; the details of within-consultation interactions; and assessment of the nature and quality of decisions 'made' therein. Such work was intended to support the operationalisation of SDM, and, subsequently, its integration into routine clinical practice. It led to the: development of models, guides, and/or protocols for practice; incorporation of these into medical education curricula; and creation of decision-specific interventions and tools, e.g., 'decision aids', to facilitate SDM (Milne et al., 2009; Elwyn et al., 2010; Stacey et al., 2011).

Notwithstanding these advances, concerns again emerged. Though arguments had been made that certain settings (e.g., paediatric and chronic care) might necessitate more complex and/or enduring partnerships (e.g., involving parents/caregivers or wider care teams) (Gabe et al., 2004; Montori et al., 2006), the assumption that SDM

took place within the boundaries of a discrete consultation, and involved two parties (a clinician and a patient) continued to be implicit in much (theoretical and empirical) work. However, in the (five) years preceding my own work, a body of research began to emerge which challenged those conceptions. That work was informed by sociological traditions and explored and threw light upon: the different ways 'significant others' may be involved in treatment decision-making (Ohlen et al., 2006); patients' use of evidence derived from their own and others' experiences when making decisions (Ziebland & Herxheimer, 2008); how decisions unfold over time (Rapley, 2008); and the ways in which decisions about self-care are grounded in everyday experiences and surroundings (the 'lifeworld') (Mol, 2009; Pickard & Rogers, 2012).

Ohlen et al. (2006) argued particularly convincingly that treatment decision-making needed to be thought of as a social, rather than an individual, process. This contention was developed further by Rapley (2008:429), who documented what he termed the 'distributed' nature of decision-making (describing how decisions could both involve a range of people, and transpire over encounters/time). These re-presentations of decision-making were powerful, and relatively widely discussed, prompting acknowledgement – in some quarters – that decision-making might leak out of, and resist containment in, the clinical encounter (Elwyn et al., 2012). Nevertheless, SDM research and literature continued for some time to be dominated by a focus upon discrete interactions between two parties (clinicians and patients). In a review of the field, published around the time I began work on my first relevant study, The Health Foundation (2012) concluded that, though clinicians and researchers had begun to acknowledge the role of family members in decision-making, important gaps in understanding remained. That report further highlighted how: 'Most studies and models of SDM do not account for inter-professional work or the need for multiple professionals to interact with patients about their decisions' (The Health Foundation, 2012:33). In short, notwithstanding the already crowded nature of the field, some significant gaps in the evidence base persisted. It was against this backdrop that the studies and papers I go on to discuss were conceived, conducted, and written up.

Introduction to the studies from which my publications arose

Shifting focus to my own work, I now introduce the three applied (qualitative) health services research studies which underpinned my submitted publications. To begin, I provide an overview, highlighting areas of similarity and difference across the studies. I then, in Tables 1-3, provide study-specific information (covering my involvement, their background, aims/objectives, design/methods and dissemination activities). In closing, I reflect briefly on how my thinking evolved as the studies progressed. I consider their findings – more specifically those documented in my publications and of relevance to decision-making – in my next, and central, chapter.

Overview: study commonalities and differences

In all three studies I was employed as the project researcher, with broadly comparable responsibilities. These included:

- refining the research design (all studies);
- securing the appropriate ethics and/or research governance permissions (at all stages for studies 1 and 2, largely for design amendments in study 3);
- handling day-to-day project administration including organising team, advisory and/or steering group meetings, facilitating patient/public involvement, and liaising with co-investigators and funders (all studies);
- collecting and managing data (all studies);
- acting as the primary analyst (all studies); and,
- writing up and disseminating the research findings (all studies).

For all three studies, funding was in place when I came into post, and my work was overseen by an academic or clinical principal investigator (PI).

Each of the three studies was concerned with the experiences of patients who (a) were receiving care within a system (the NHS) in which SDM was aspired to (Department of Health, 2010) and (b) were confronted with some sort of choice(s) about their treatment and/or management (including self-care). However, the studies varied in how directly and explicitly they focussed on patients' involvement in decision-making; this was not always their primary focus. Indeed, in the case of study 2, it was very much a subsidiary concern. Nevertheless, as I shall go on to

demonstrate in the following chapter, all progressed my thinking about, and understanding of, health-related decision-making in some meaningful way.

The patients of interest, and the decisions they faced, differed across the studies in potentially important ways. Studies 1 and 3 explored the experiences of patients aged 16-24/25 years; patients participating in study 2 ranged more widely in age (25-77 years). Patients in studies 1 and 2 were contending with chronic, and largely established, disease. In contrast, those in study 3 were experiencing more acute, newly diagnosed, disease. Hence, patient-participants were confronted with markedly different decisions. These were whether to: try an alternative, new(er) form of medication with a distinctive profile of potential effects and side effects (study 1); engage with patient information materials and adopt non-pharmaceutical approaches to symptom management (study 2); or, opt for treatment administered through a clinical trial (study 3).

All three studies built on earlier, sociologically-informed work on decision-making (discussed above) to which I was introduced by my PI and colleagues at Newcastle University. Each was conceived and conducted with an appreciation that there might be influences on decision-making not visible in clinical encounters – and, moreover, that decision-making might be affected by factors not disclosed by, or not apparent to, patients themselves. This awareness informed both research questions and design choices (e.g., sampling, topic guide content). Hence, in all three studies data were collected not only from patients, but also from health professionals. Additionally, in my first and third studies, data were collected from 'trusted others', allowing for the possibility that their perspectives might enhance understanding of patients' experiences. All three studies employed semi-structured interviews as their principal data collection method; however, they differed significantly in their scale (number and range of participants). Study 1 additionally gathered data via recordings of clinical consultations and a series of focus groups (with young patients, parents/caregivers, and professionals). In both relative and absolute terms study 2 was modestly funded, constraining research activity (data collection and analysis) and the range of study outputs.

Thus, the studies demonstrated both similarities and differences: further study-specific information can be found in the following Tables (1-3).

Study 1: How should we educate young people with inflammatory arthritis to help them make decisions about biologic therapies?	
Involvement in study planning, design and delivery	I was engaged in this research from 2012-2014. Funding had already been secured, from Arthritis Research UK, but the research design and instruments (e.g., topic guides) remained to be finalised. Ethics and governance permissions were also needed. I refined the project plan, drafted the instruments, and prepared other key materials (e.g., information sheets, consent forms, and ethics applications). Subsequently I played an instrumental role in decisions about research design changes (e.g., modification of sampling and recruitment strategies, refinement of topic guides). I collected all data, and conducted the bulk of the analytical work. I received support and oversight throughout from the grant holder/PI, Dr. (now Prof.) Tim Rapley, a medical sociologist.
Background to the study	The term inflammatory arthritis (IA) encompasses a number of systemic auto-immune conditions including rheumatoid arthritis (RA), psoriatic arthritis (PsA), ankylosing spondylitis (AS) and juvenile idiopathic arthritis (JIA). Whilst typically occurring in older adults, these conditions also affect children and young people. Management of IA has evolved significantly, with one important development prior to the study being the increasing use of biologic therapies (treatments targeting immune components of disease). These had demonstrated many short-term benefits, but their longer-term effects remained uncertain. Hence, patients offered biologics needed to think carefully about their potential benefits and (hypothetical) risks. Health professionals needed to help people do this by providing counselling appropriate to their needs: we surmised younger patients might require and/or value different sorts of information and support to older adults.
Aim(s) and objectives	**Investigate the experiences and perspectives of 16-25 year-olds diagnosed with IA, in order to identify particular educational needs.** Explore experiences of decision-making about biologic therapies; describe information-exchange in consultations; understand the influence of 'trusted others'; identify values regarding risks and benefits; and describe decision-making processes.
Design and methods	This study used semi-structured interviews, recordings of clinical consultations, and focus groups to elicit the perspectives of young patients, trusted others, and health professionals. I identified and recruited patients via the rheumatology services (adult, young adult, and/or adolescent clinics) of three NHS Hospital Trusts. Patients nominated and approached trusted other interviewees. Sampling of patient and professional interviewees was purposive, seeking variation in: patients' (n=25) demographic characteristics, diagnosis, and treatment status; and, health professionals' (n=6) roles and responsibilities. I interviewed patients up to three times (where their treatment status changed). In addition, four consultations about biologic therapies were recorded. I also convened four focus groups: two involving young people; one with trusted others; and one with health professionals. Recordings were transcribed verbatim for analysis. In this I used techniques derived from grounded theory (open and focussed coding, constant comparison, deviant case analysis, memoing and mapping) and received support and oversight from the PI. I discussed findings and their implications with the wider team and advisory group (which included both patients and professionals).
Dissemination of study findings	Key findings from this study were published in *Rheumatology* (journal of the British Society for Rheumatology (BSR)) (**Paper 1**) and in *Arthritis Care & Research* (journal of the American College of Rheumatology and the Association of Rheumatology Health Professionals) (**Paper 2**). I also gave talks to patient support groups and professional audiences, including presenting at: the British Society for Paediatric Rheumatology Research Day 2013; and Rheumatology 2014 (BSR Annual Conference).

Table 1

Study 2: The practical management of fatigue by people with arthritis: the impact of Arthritis Research UK written information.	
Involvement in study planning, design and delivery	I conducted this research in 2014-2015. Again funding from Arthritis Research UK was in place when I transitioned to the project. Similarly, however, certain aspects of the research plan remained to be finalised, instruments and other study documents needed to be prepared, and appropriate research permissions had to be secured. I completed these tasks before beginning data collection and analysis, for both of which I had primary responsibility. Support and oversight was provided by the PI, Dr. Ben Thompson, a clinician with interests in qualitative research.
Background to the study	Fatigue (extreme and often unexpected tiredness) is a common symptom of arthritis and associated conditions, which can substantially reduce quality of life. When the project was undertaken effective treatments remained to be found, though there were ways in which fatigue and its impact could be managed. It was surmised that understanding how people dealt with fatigue, and if/how information such as the 'Fatigue and arthritis' booklet developed by Arthritis Research UK could help them with this, might enable professionals to support patients better.
Aim(s) and objectives	**Investigate people's approaches to managing fatigue and the impact Arthritis Research UK's booklet has upon them.** Explore how people with three types of arthritis (RA, AS and primary Sjogren's syndrome (pSS)) make and implement decisions about the management of fatigue and attempt to reduce its impact on their life; determine how Arthritis Research UK's published material connects with, and influences, patients' lived experiences of fatigue; and evaluate the 'Fatigue and arthritis' booklet, as a foundation for the revision and further development of Arthritis Research UK's patient information materials.
Design and methods	This qualitative study had a longitudinal design, involving 'pre-' and 'post-' intervention (booklet) interviews with patients (n=13); and a focus group comprising 'expert' patients (n=2) and professionals (n=6). I identified and recruited patient-participants from one NHS Hospital Trust, seeking variety in diagnosis (RA, AS, pSS), fatigue severity, age, gender, general health, life circumstances and demands. I interviewed patients at two time points, roughly 4-6 weeks apart. Their perspectives were presented to focus group members, and implications for the future development and use of the booklet discussed. The interviews and focus group were audio-recorded and transcribed. I analysed the data using techniques derived from grounded theory (e.g., constant comparison), seeking to identify patterns (recurring issues or themes) and develop explanations for these.
Dissemination of study findings	I prepared one publication (**Paper 3**) providing an overview of study findings. This appeared in *Musculoskeletal Care* (a journal for health professionals involved in the care for people with musculoskeletal conditions). In addition, I prepared a poster for presentation at the American College of Rheumatology's annual (2015) conference, and contributed to formal and informal discussions about fatigue and its management.

Table 2

Study 3: Barriers and facilitators to participation in cancer trials amongst teenagers and young adults (TYA).	
Involvement in study planning, design and delivery	I worked on this study from 2017-2019, coming into post after funding (from the Chief Scientist's Office) and initial ethics permissions had been secured. Considerable work remained (which I undertook) to ensure appropriate governance permissions and arrangements were in place to recruit from health boards across Scotland. The study's emergent design provided scope for me to shape it further: I revised the various topic guides and was instrumental in modifying sampling plans (to enable collection of data from a wider range of professionals). Support and oversight for my work was provided by the PIs, Prof. Julia Lawton (a medical sociologist) and Dr. Angela Jesudason (a clinician).
Background to the study	Trials are pivotal to improving clinical care, but when the study was conceived, only 10 per cent of TYA with cancer in Scotland took part in one.
Aim(s) and objectives	**Understand the reasons for low levels of trial enrolment amongst TYA with cancer in Scotland, and how levels of participation might be improved.** Explore TYA, caregiver, and health professionals' experiences of and views on being recruited, or recruiting, to trials (including factors affecting whether health professionals approach TYA about trial participation) and their respective views on how they, and others, might influence TYA(s)' decision-making about trial enrolment, as well as how they think trial participation by TYA with cancer might be improved.
Design and methods	This qualitative study involved in-depth interviews with: TYA diagnosed with cancer whilst aged 16-24 years (n=18); caregivers (n=15); and health professionals (n=35). I identified and recruited TYA from paediatric and adult cancer centres across Scotland, seeking to sample purposively to achieve variation in age, gender, diagnosis, and place of care. Participating TYA then nominated caregiver interviewees. I identified potential health professional interviewees with the help of clinical members of the research team, advisory group members, and other colleagues. Initially I focussed on recruiting direct care professionals (seeking variation by role, sub-specialty, service, hospital, and health board). Then, in light of my preliminary analysis, which suggested significant structural barriers to trial participation, I made the case for and secured the necessary permissions to recruit an additional sub-set of professionals involved with the facilitation and/or delivery of clinical trials in Scotland. All interviews were audio-recorded and transcribed verbatim for analysis. That analysis, which I undertook in conjunction with Prof. Julia Lawton, involved 'line-by-line' and more focussed coding, and 'constant comparison', to arrive at descriptive themes. I sought feedback on the emerging findings and their implications for policy and practice from the wider research team and study advisory panel members (health professionals and TYA with cancer).
Dissemination of study findings	I prepared four papers (**Papers 4-7**) which were published in the following peer-reviewed journals: *Trials* (journal publishing articles on general trial methodology and research into trial processes); *BMC Health Services Research* (journal publishing articles on all aspects of health services research); *Health Expectations* (an interdisciplinary journal publishing papers on patient and public involvement in health care, policy and research); and *European Journal of Cancer Care* (journal publishing papers on issues affecting the care of cancer patients). I also wrote a lay report, published on the funder's website.

Table 3

My evolving orientation to the topic of health-related decision-making

Prior to beginning work on study 1 I knew little, and indeed had thought little, about health-related decision-making. My early experiences on that study had a powerful impact on me, and directly and indirectly informed my work going forward. In particular, emergent findings from study 1 highlighted the complex, messy and varied realities of health-related decision-making, inviting a degree of scepticism about the adequacy of traditional models of SDM. That scepticism led me (in both that first and subsequent studies) to attend very closely to matters such as:

- how people other than patients and clinicians are involved in and/or influence decision-making;
- the different sorts of information people draw on, and how, when confronted with health-related decisions;
- the influence of direct, personal (corporeal and/or lived) experience, when people formulate and/or evaluate such decisions; and
- linked closely to the topic above, the contingencies of any decisions made (and their implications for sustained implementation).

These interests have informed my sampling decisions, the content of my topic guides, and the areas of my datasets prioritised by me for analysis and ultimately reporting (see **Papers 1, 2, 3, 5** and **6**).

My involvement with study 3 prompted a further, important shift in perspective, imbuing me with a strong sense that whilst the patient perspective is clearly important, it may not alone be enough for us to understand decision-making experiences fully. To expand, data gathered from direct care professionals in the initial phase of that project strongly suggested that structural factors had a far more important explanatory role (in decision-making about cancer trial participation) than colleagues or I had appreciated at outset. This discovery prompted me to secure permission to reformulate the study's sampling and data collection strategy, to enable further exploration of how those wider factors shaped the decisions patients were invited to make. In **Papers 4** and **7** I documented how this more 'upstream' approach could extend understanding of decision-making in very important ways.

Fundamentally, my involvement with these three studies left me with the conviction that to reach a comprehensive understanding of patients' experiences of decision-

making *within* clinical consultations, it is likely to be important also to consider and take account of what goes on around them. That belief has only been strengthened by my work on this critical review, which has provided a vehicle for revisiting previous research and consolidating key findings.

What follows: outline of the critical review

In the following chapter I present key findings from my seven submitted publications of salience to the topic of health-related decision-making. These take the form of a series of themes, arrived at via a process of qualitative synthesis, which provide the building blocks for the proposition, or line-of-argument, that health-related decision-making happens in and is shaped by its personal, social and health service contexts. Subsequently, I consider how my findings complement and challenge the wider literature. I argue that, in highlighting the role of context, my papers and this review provide insights that enhance and extend the historic emphasis in SDM scholarship on what goes on within clinical encounters. I also note where other, diverse literatures, might usefully inform thinking. In closing, I contend that appreciation of context is crucial both to understanding and, moreover, to facilitating, patients' involvement in health-related decision-making.

SYNTHESIS OF FINDINGS FROM MY PUBLICATIONS

In this chapter I present salient findings from my publications and demonstrate how, collectively, they constitute a coherent body of knowledge which extends the existing literature on, and understandings of, health-related decision-making. Though, as already noted, my studies varied in the extent to which they directly addressed the topic of decision-making, I judged all seven papers to have the potential to contribute something. This was confirmed through a process of qualitative synthesis (detailed below) via which I developed the proposition, or line-of-argument, that health-related decision-making happens in and is shaped by its personal, social and health service contexts.

Approach to the synthesis

Why undertake qualitative synthesis and what does it seek to do?

Though efforts to synthesise findings from qualitative studies date back to the late 1980s (Noblit & Hare, 1988), it was another decade before the idea of bringing together and seeking to extend the findings of discrete studies gained widespread attention. Such work was initially highly controversial – for example, Sandelowski et al. noted how, in light of qualitative research's emphasis on idiographic knowledge, it seemed inherently problematic to attempt to sum up or generalize from the findings of distinct studies:

> *'To summarize qualitative findings is to destroy the integrity of the individual projects… thin out the desired thickness of particulars… and, ultimately, to lose the vitality, viscerality, and vicariism of the human experiences represented in the original studies.'* (Sandelowski et al., 1997:366)

However, as these authors themselves countered, and others similarly recognised, failure to try to link and build upon the body of existing qualitative work risked holding the tradition back – both in respect of the production of more sophisticated understandings, and in its ability to offer useful, practical knowledge (Sandelowski et al., 1997; Campbell, 2003).

As interest in 'evidence-based' medicine (and policy) grew, further attention was given to whether, and how, qualitative synthesis might – like meta-analysis of quantitative studies – usefully inform policy and practice. Questions and concerns started to focus on how to do qualitative synthesis 'right' (Thomas & Harden, 2008),

rather than whether to attempt it at all. Concerns emerged that quantitative approaches, concerned primarily with aggregation, might provide a template for qualitative synthesis, with critics suggesting that qualitative synthesis could, and indeed should, not solely summarise, but seek, in addition, to deliver more advanced conceptual understanding (Campbell et al., 2003). Other authors argued that this characteristic of 'going beyond' the findings of contributing studies was what ultimately distinguished synthesis from traditional (literature) reviews (Britten et al., 2002; Thomas & Harden, 2008; Lee et al., 2015). This meant not only drawing out concepts or themes from synthesised work, but also developing new or 'synthetic' ones, which captured and explained interesting aspects of the 'data'. The nature and function of such constructs was described by Dixon-Woods et al. as:

> *'grounded in the evidence, but result(ing) from an interpretation of that evidence, and allow(ing) the possibility of several disparate aspects of a phenomenon to be unified in a more useful and explanatory way.'* (Dixon-Woods, 2005:17)

Debates have continued, and as a relatively young enterprise, qualitative synthesis continues to raise interesting epistemological and methodological questions. However, within applied health services research its value is now widely recognised, and it is largely accepted that qualitative synthesis can serve a range of very useful purposes, including but not limited to those described by Kimbell et al.:

> *'Synthesising bodies of qualitative literature can help clarify understanding of a phenomenon, identify gaps and ambiguities in the existing literature, and inform decision-making by policymakers and healthcare practitioners'* (Kimbell et al., 2021:2).

How was this particular qualitative synthesis undertaken?

A number of approaches to qualitative synthesis have emerged. Though these differ in some important ways (discussed below), most involve some common preliminary activities. These include systematic (though not necessarily exhaustive) searches of the literature to identify publications meeting pre-specified criteria, and examination of such publications for the purposes of screening and appraisal. The work I report diverged from that norm in the following ways and for the following reasons. Firstly, a systematic search for relevant studies was not undertaken as the goal of the work was to synthesise pertinent findings from a circumscribed set of publications meeting

rather unusual criteria (personal authorship). Secondly, as the publications were already familiar, rather than undertaking screening and appraisal in the conventional sense, they were examined to confirm their potential to provide insights germane to the topic of interest, and assess whether there was sufficient overlap in their concerns to support the drawing out of themes (i.e., essentially to evaluate the feasibility of undertaking a synthesis). Though the synthesised papers differed in their focus and the extent to which their content pertained directly to decision-making, I judged all to have the potential to contribute something. As the body of work was not so large as to require reduction at an early stage to render it manageable, no material from the publications was bracketed off or discounted before its potential to contribute to the synthesis was fully explored.

The following paragraphs detail how I selected an approach for the core (analytical) work of synthesis. As noted above, various approaches have been proposed; indeed, Booth et al. (2016) identified more than 20. These include meta-ethnography, (qualitative) 'meta' -study, -synthesis, -interpretation and -analysis, as well as critical interpretive synthesis, narrative synthesis, realist synthesis, framework synthesis, and thematic synthesis (Noblit & Hare, 1988; Sandelowski et al., 1997; Paterson et al., 2001; Pawson, 2002; Pawson et al., 2004; Thorne et al., 2004; Dixon-Woods et al., 2006; Bondas & Hall, 2007; Oliver et al., 2008; Thomas & Harden, 2008; Dixon-Woods, 2011; Campbell et al., 2011; Carroll et al., 2013; Booth et al., 2016).

Not all these approaches have been clearly operationalised and/or are easily distinguished from each other: reviewing the field, Campbell et al. (2011) noted that different terms had been used to describe very similar approaches, and the same terms used to describe quite different approaches. Over the last decade certain approaches (or terms) appear to have fallen into disuse, whilst others appear to have become more prominent. These (those to which I gave serious consideration) are meta-ethnography, realist review, framework and thematic syntheses (and their derivatives). They demonstrate some important differences: in their emphasis (aggregation or interpretation); methods; the sorts of literature they have been (successfully) applied to; and the nature of their products (Thorne et al., 2004).

Meta-ethnography, for example, was initially devised as a means of synthesising a very small number of relatively lengthy, theoretically-developed and closely-related monographs (Noblit & Hare, 1988). Campbell et al. (2003) subsequently explored its application to a different and larger group of studies (specifically, a set of phenomenological papers, all written for social science audiences). Whilst they judged this exercise productive, they cautioned it remained unclear whether more diverse sorts of studies and publications might be suited to meta-ethnography. Subsequently attempting to use this approach to synthesise findings from a larger and more methodologically varied group of studies, Dixon-Woods et al. (2006) ended up modifying it to the point where they judged themselves to have developed a new method (Critical Interpretive Synthesis, or CIS). Though Campbell et al. (2011) questioned whether CIS warranted the status of a distinct method, it too has been relatively widely used/cited in recent years. Evaluating meta-ethnography, Campbell et al. (2011) opined that whilst the approach had promise, the associated demands were considerable, and new insights not guaranteed. These conclusions are consistent with my own and colleagues' experiences of attempting to apply meta-ethnographic techniques to more applied literature (peer-reviewed papers, written for a clinical/health services audience) (Lee et al., 2015). Those experiences led me to suspect that seeking to identify and translate 'metaphors' (key concepts) between and across a small number of succinct and applied research papers such as those in my portfolio was likely to be at best unconvincing, and at worst futile. As colleagues and I noted:

> *'the best efforts of the meta-ethnographer may be frustrated by the nature of the accounts being synthesised.'* (Lee et al., 2015: 344)

Recent years have seen the emergence and increasingly widespread application of less interpretive approaches, adapted from primary research and oriented towards the production of findings for use by practitioners and/or policy makers. These include realist review/synthesis, framework synthesis, and thematic synthesis. The first of these, realist review/synthesis is an approach to reviewing research evidence on complex social interventions, underpinned by a generative theory of causation and aiming to explain how, when and why such interventions do or do not work (Pawson, 2002; Pawson et al., 2004; Pawson et al., 2005; Wong, 2013). Wong (2013) noted how the approach is well suited to some topics (associated with

understanding how programmes work) but less useful for others. I quickly discounted this approach on grounds of poor fit with both my goals and the corpus of literature available for the synthesis. Framework synthesis, on the other hand, appeared reasonably well suited. Dixon-Woods (2011) noted how many of the characteristics of framework analysis making that approach popular in applied primary research (clear steps, transparency of activity, and a focus on stakeholders' interests) were also present in framework synthesis. However, I was deterred by its emphasis on *a priori* concerns (or in the case of 'best fit' framework synthesis, a pre-existing model or theory) which has the potential 'to suppress (the) interpretive creativity' needed to go beyond summarising the findings of contributing studies (Dixon-Woods, 20011:2).

Ultimately the approach I chose to adopt was thematic synthesis; a clearly operationalised, accessible, relatively pragmatic, but still essentially inductive method. I deemed this well-suited to working towards the goals I had in mind, and with the literature available to me. The approach was originally developed to help address policy questions which could not be answered satisfactorily on the basis of synthesising quantitative studies alone (Thomas & Harden, 2008). However, it has been widely used since to explore patients' (and others') perspectives and experiences. Indeed this approach was recently and successfully applied by two groups of colleagues (Cavers et al., 2019; Kimbell et al., 2021), adding to my confidence that it was suitable and workable for synthesising a relatively small body of applied health services research publications.

Thomas & Harden (2008) asserted that their approach encourages closeness to the results of the contributing studies, and that, whilst capable of generating new analytical concepts, thematic categories, and propositions, it makes the process of synthesis highly transparent. They identified three core stages of work as involved in thematic synthesis:

- free/open line-by-line coding (of study results/findings);
- organisation and grouping of these codes, through comparison of similarities and differences, into (relatively concrete) descriptive themes; and
- development of more abstract thematic categories or 'analytic themes', via the close consideration of patterns and relationships in and between the

descriptive themes, with a view to achieving a new level of understanding, expressed in an overarching 'proposition'.

The parallels with thematic analysis – 'a method for identifying, analysing and reporting patterns (themes) within [primary] data' (Braun & Clarke, 2006:79) are marked. Similarly, the analytical work of thematic synthesis can be undertaken using highlighters, sticky notes and index cards, facilitated by the use of analytical software, and conducted by a lone researcher or a team (generally judged to enhance rigour). I used several of these tools, initially coding and grouping codes into descriptive themes (and developing these in turn into provisional thematic categories) on paper, and subsequently reviewing the data and refining codes, themes and categories with the aid of NVivo 11 (QSR International, Doncaster, Australia). As Thomas & Harden (2008) have argued, observing these processes ensures a clear and confident interpretative trail can be followed – from codes, through descriptive sub-themes/themes, to analytic themes or categories, and overarching proposition(s). This trail can be developed and presented using tables and/or thematic maps (Braun & Clark, 2006).

Findings from the synthesis

My synthesis led, ultimately, to the proposition that ***health-related decision-making happens in and is shaped by context***. It highlighted the importance of three particular kinds of context – the personal, social, and (health) service contexts – each associated with an analytic theme. These themes are: *experiencing a state of dis-ease*; *a source of support and influence;* and, *creating a menu and climate for decision-making.* The three analytic themes are underpinned by nine descriptive themes. Below, I describe each of my analytic and descriptive themes in turn, offering relevant illustrations and noting how they link to cross-cutting instrumental themes (i.e., mechanisms via which decision-making is influenced). Figure 1 (overleaf) provides an overview of the different themes and their relationships with each other. Appendix 1 contains additional thematic maps (Figures 2-4) illustrating the development of the three analytic themes.

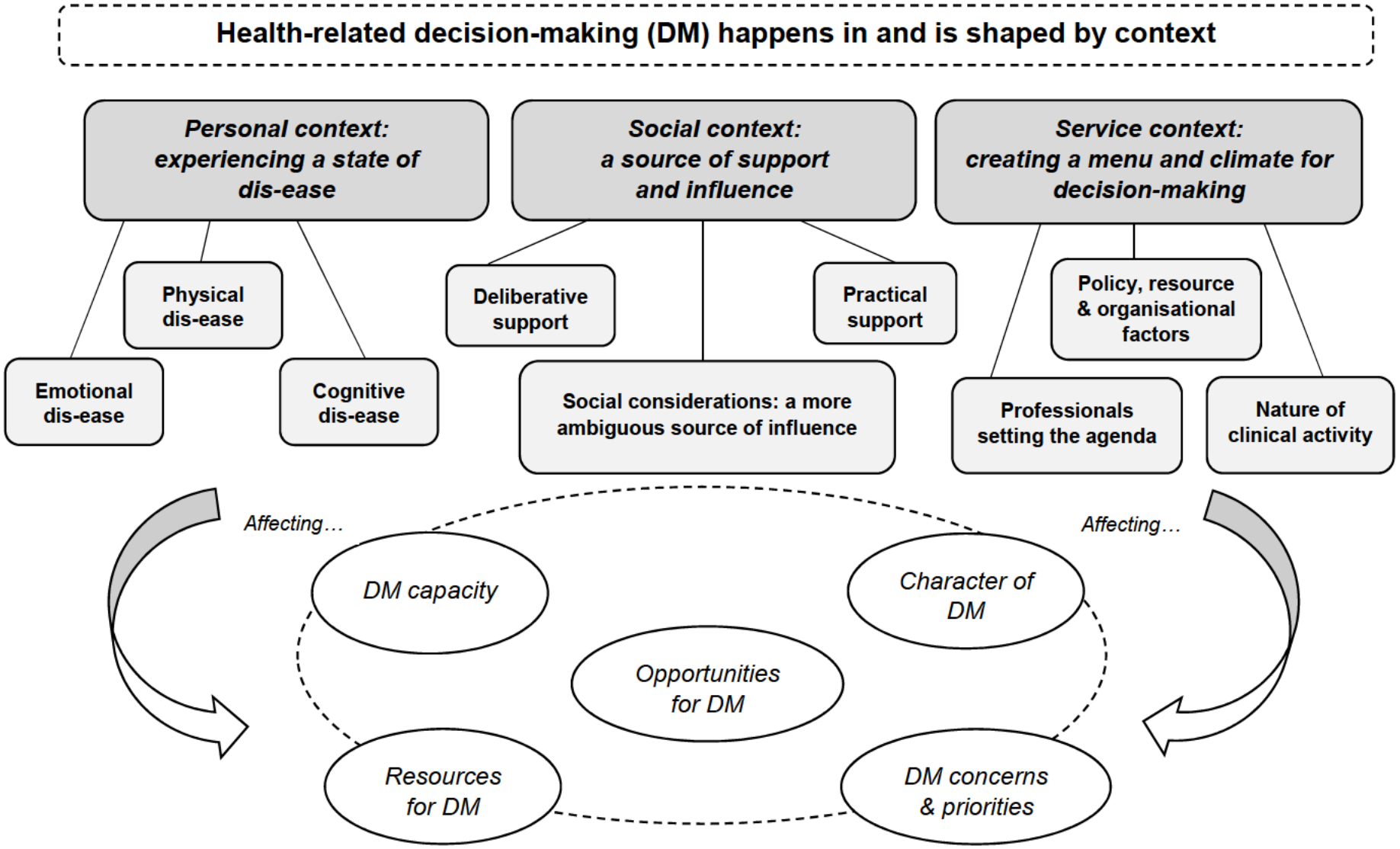

Figure 1: Map of overarching analytic themes, principal descriptive themes, and instrumental themes

Analytic theme 1: Personal context – experiencing a state of dis-ease

Firstly, my synthesis highlighted the personal, embodied context in which decision-making took place, and how often this was characterised by experiences of physical, emotional, and/or cognitive 'dis-ease'. I employ this term (dis-ease) deliberately, to connote un- or lack of ease – a sense of discomfort, being 'off-kilter', or out of balance – rather than illness or pathology *per se*. I developed the concept inductively, only subsequently becoming aware of the term's use by others (e.g., Antonovsky, 1979; Vinje et al, 2017). Below, I detail how each of the three identified forms of dis-ease (physical, emotional and cognitive) provided a backdrop to patients' decision-making, and shaped their decision processes (and choices) in different and potentially important ways.

Experiencing physical dis-ease

My synthesis underscored the ubiquity of some level of physical dis-ease, in particular in the form of pain or fatigue. Participants in all three studies described physical dis-ease arising: initially, from symptoms; but also (in studies 1 and 3) from clinical interventions (e.g., investigations, symptom management and/or treatment). Drilling down further, I noted variation: my synthesis illuminated some distinct ways physical dis-ease could inform decision-making.

To begin, I observed how physical dis-ease arising from symptoms could create a profoundly challenging backdrop for decision-making. Findings from study 3 showed how, for many patient-participants, physical dis-ease associated with symptoms had escalated dramatically in the lead-up to their diagnosis (with cancer). Many described themselves (or were described by others) as having felt 'deeply' or acutely unwell by the time they were faced with decisions about treatment; indeed some reported having been in-patients by that time. In **Paper 5** I described in some detail how severe pain, exhaustion, and other disabling symptoms presented barriers to these patients engaging with (complex) disease and treatment-related information. For example, one patient's mother explained:

> *'she was exhausted and I think a lot of the time (daughter) was leaning on me zonked out … she didn't really, really understand the full implications of what it (treatment) was going to entail'* (**Paper 5**, p6)

Findings from that study additionally indicated how clinical interventions (investigations or interim treatment) could contribute further to experiences of

physical dis-ease. For example, I described (in **Paper 5**) how several TYA reported having undergone investigative surgery shortly before receiving their diagnosis. Some recalled having been under the influence of powerful analgesics at diagnosis and first discussion of treatment, with this further impairing their capacity to absorb and process information:

> *'(I was) so dosed up on morphine that I had no idea what that (diagnosis) meant'* (**Paper 5**, p4)

Both TYA and their parents suggested that physical dis-ease – originating from symptoms or clinical interventions – had deleterious consequences for whether and how effectively TYA were able to make use of information salient to treatment decision-making. Indeed, whilst recognising that decisions needed to be made and actioned promptly (to prevent further, potentially life-threatening, deterioration) some parents expressed profound concerns about TYA(s)' capacity to make informed decisions under such conditions.

Findings from my earlier studies offered something of a contrast, suggesting physical dis-ease might also affect decision-making in other – less problematic – ways. Participants in those studies described experiencing, and struggling day-to-day, with physical dis-ease associated with common symptoms of rheumatic diseases (pain, joint swelling, stiffness, and fatigue). They talked of chronic discomfort and disability, describing wide-ranging impacts on their mobility, domestic, social and vocational lives:

> *'Socializing, just doing things that you want to do, are rather harder, or get put on hold, because you're tired all the time'* (**Paper 3**, p233)

However, these interviewees' accounts did not suggest that their decision-making capacity was routinely compromised by physical dis-ease arising from symptoms.

Instead, patient interviewees highlighted the fluctuating nature of their symptoms, and how they had 'good' days and 'bad' (see **Paper 2**). Hence, they suggested, whilst physical dis-ease (e.g., pain) could disrupt decision-making at times, its effects were less marked at others. And, as their conditions did not, in the short-term, present a threat to life, taking time to reflect on, investigate, and even reconsider treatment proposals was generally an option. Notably, though there were exceptions, these patients appeared both physically able and motivated to engage with detailed

written information about their condition and its treatment, as a basis for decision-making. Physical dis-ease associated with symptoms often appeared to be an incentive for, rather than a barrier to, discussion and deliberation about treatment options.

Moreover, by virtue of having relatively established (chronic) conditions, patients in studies 1 and 2 often had experience of a variety of treatments, including some with challenging side effects, i.e., themselves causing physical dis-ease. Accounts suggested that such prior experiences (of intervention-related physical dis-ease) could serve as an important influence on, and, indeed, a resource for, treatment decisions. For example, interviewees revealed an awareness that certain medications might bring about new forms of dis-ease, with associated psycho-social costs. In **Paper 2**, I described how people reflected upon the challenging side effects and social impacts of some prior treatments, such as methotrexate:

> *'Every Monday, you could guarantee that I wasn't at school … 'cause I was still sick from the medicine'* (**Paper 2**, p1291)

These interviewees explained how prior experiences shaped their concerns and priorities when confronted with decisions about new or alternative regimens. They hoped for both relief from symptoms and reduced treatment burden, to improve the quality, scope, and predictability of their lives. In essence, one interviewee explained, they wanted treatment that made life 'easier … rather than harder' (**Paper 2**, p1292): where this was not the case, then decisions might be revoked. Essentially, these interviewees framed treatment decisions as provisional: they would 'try it and see' (**Paper 2**, p1292).

Hence, my synthesis showed physical dis-ease to be a ubiquitous yet differentiated experience. In its acute form(s) it could substantially impede informed decision-making, whilst chronic variants might promote engagement with treatment information, enable recourse to experiential knowledge, and encourage experimentation.

Experiencing emotional dis-ease

My synthesis similarly found emotional dis-ease to be a common feature of patients' experiences. Across the studies, participants related experiencing various complex and challenging emotions (fear, anxiety, shock, confusion, frustration, guilt, sadness

and grief). As I describe below, emotional dis-ease manifested in different ways, but appeared a consistent and often disruptive companion to decision-making.

'Acute' (i.e., severe or intense) emotional dis-ease characterised the accounts of participants in study 3. I described in **Paper 5** how TYA patients (and their parents) recalled experiencing a 'rush of emotion' in response to the news that they (or their child) had cancer. These interviewees reported feeling shock, extreme distress and fear (plus occasionally also relief at the prospect of treatment):

> *'I'm not going to lie … I was a wee bit stunned and then I was really upset, we were all crying and then I was a bit like, what's going to happen … but then part of me, in a weird way was like, this is gonna take the pain away. Like the pain was that bad'* (**Paper 5**, p4)

Interviewees suggested a range of factors precipitated the emotions described: the unexpectedness of a cancer diagnosis; being confronted with the possibility of death; and (finally) getting an explanation for profound physical dis-ease.

With regard to decision-making, TYA and their parents explained how, as with physical dis-ease, emotional dis-ease could impede engagement with important information:

> *'like chemo and that… I was just too sad to read them (leaflets)… Yeah, just, I'd just end up crying'* (**Paper 5**, p6)

I also noted in **Paper 5** how some TYA reported trying to minimise emotional dis-ease by actively avoiding potentially distressing information (for example, on disease prognosis). Though this strategy may have helped them cope, parents were again concerned about the implications for informed decision-making.

A different sort of emotional dis-ease (lower level, but chronic) was revealed by participants in studies 1 and 2, with anxiety being particularly prominent in their accounts. Interviewees described feeling worried about the cause of symptoms, future treatment experiences, and social impacts. Illustrating the first of these concerns, patients in study 2 (**Paper 3**) disclosed anxieties about the origins and significance of their fatigue. They also shared concerns about how their fatigue-related behaviours were viewed by others. Participants in study 1 (**Paper 2**) talked more extensively about anxieties regarding treatment, with some explaining how even small changes to their regimes could trigger powerful emotions. Typically these

interviewees had prior experience of both treatment that was limited in its effectiveness, and treatment that was very difficult to tolerate:

> *'The methotrexate worked for a little bit and then it stopped'* (**Paper 2**, p1292)

> *'The look of it, the smell of it, the very thought of it made me shake'* (**Paper 2**, p1291)

The former experience left people wary of disappointment, whilst the latter led to anxieties about the discomfort or physical dis-ease new treatments might entail. Hence, unfamiliar treatments were a source of considerable anxiety for these young people.

Again (i.e., in its chronic as well as acute form) emotional dis-ease could lead to information avoidance. TYA in study 1 were focussed on living as well as possible with their disease, and were concerned primarily with information relevant to their immediate, short-term experiences. Unlike their peers in study 3, who prioritised longer-term outcomes, they did not look closely at the potential future consequences of treatment. Indeed, in **Paper 2** I reported how some TYA described having made an active choice not to attend to the longer-term risks of treatments effective in the short-term:

> *'If something bad happens, I'll cross that bridge when I come to it. For now, it's just keeping me normal. I know that sounds a bit reckless, and I don't mean it like that, but like, I can't worry about what, you know, what would happen'* (**Paper 2**, p1292)

Once more, this selective approach to information was a concern to some parents.

Thus, my synthesis showed how emotional dis-ease was widespread, and, whether manifesting in 'acute' or 'chronic' form, could affect (discourage) patients' engagement with information considered by others to be salient to treatment decision-making.

Experiencing cognitive dis-ease

Finally, my synthesis suggested a third form of dis-ease, relating to cognition or mental processes, was similarly widespread. This too appeared to manifest in different ways – including as uncertainty and overload – with potentially problematic consequences for decision-making.

Firstly, considering cognitive dis-ease in the shape of uncertainty, my synthesis drew attention to instances where patients lacked clarity about the meaning and significance of information – both somatic information and that received from external sources. For instance, findings from studies 2 and 3 revealed how people struggled to make sense of non-specific symptoms, in particular fatigue. Indeed, I reported in **Paper 3** how some people were surprised to discover, via the research, that fatigue might even be a 'symptom' of rheumatic disease:

> *'It hadn't occurred to me that it might be part and parcel of the condition'* (**Paper 3**, p233)

Similarly, I noted in **Paper 6** how some TYA diagnosed with cancer described having experienced but been perplexed by fatigue, sometimes waiting long periods before discussing it with a health professional.

My synthesis revealed how difficulties interpreting symptoms had important practical and decision-relevant consequences. In the examples above, uncertainty as to the significance of symptoms left people doubtful and/or confused about the appropriate course of action: hence, they struggled to make and/or action decisions about help-seeking. Participants in study 2 did not think – or feel able to – raise fatigue in consultations with their rheumatologist, and TYA with cancer (study 3) did not prioritise getting medical advice until more clear-cut or problematic symptoms developed.

Study 3 also offered examples of uncertainty around the meaning of information received from external sources e.g., health professionals. In **Paper 6** I reported how TYA patients described the language of cancer, and indeed healthcare, as unfamiliar, acknowledging in hindsight that they had not understood all the information provided. Again this had consequences. For instance, some TYA explained how lack of clarity regarding the purpose of investigations led to them attending pivotal appointments alone (e.g., consultations in which their cancer diagnosis was disclosed):

> *'I went by myself (to get the results of biopsy), because, like I say, I wasn't expecting… [Interviewer: That sort of news?] Yeah'* (**Paper 6**, p6)

Whilst my synthesis indicated that uncertainty arising from lack of knowledge, or ambiguous information, could lead to poor choices, it also suggested that excess of

information could cause difficulties. Findings from study 3, in particular, pointed to a further form of cognitive dis-ease, which I have termed 'overload'. I noted in **Paper 5** the immense amount of information (on diagnosis, treatment, and care pathways) offered to TYA with cancer following diagnosis. Many TYA in that study talked of feeling overwhelmed by the volume and complexity of information given to them over a short period of time and whilst they were experiencing physical and/or emotional difficulties (dis-ease):

> *'the bombarding of information in the first week. It's a lot of information to take in'* (**Paper 5**, p7)

TYA further described how this sense of informational overload prompted different reactions relevant to decision-making about treatment. Some, for example, reported looking to health professionals for a steer where any important decisions had to be made (see **Paper 5**). Others explained how they coped by attending selectively to the information provided. For example, in **Paper 5** I reported how some TYA with cancer portrayed themselves as almost exclusively concerned with, and focussed upon, survival and the longer-term:

> *'the things that do matter to me are the long-term side-effects, what's my life going to look like in five years, 10 years'* (**Paper 5**, p6)

They chose not to attend to the short-term side effects of the treatments they faced; some were consequently shocked by their 'pervasive and brutal impact' (**Paper 6**, p7).

Thus, my synthesis drew out examples of cognitive dis-ease manifesting as uncertainty and overload. It showed how uncertainty could have important practical implications, whilst overload might prompt the adoption of pragmatic, coping strategies. Both such consequences could, potentially, affect experiences and outcomes of decision-making.

Analytic theme 2: Social context – a source of support and influence

Next, my synthesis highlighted the importance of patients' social contexts or worlds, revealing a variety of ways in which people other than health professionals provided deliberative and/or practical support for decision-making. It also, however, called attention to the potential for social considerations to influence decision-making in more ambiguous (i.e., less unequivocally positive) ways.

Deliberative support: the role and importance of 'infomediaries'

All three studies called attention to the ways people other than professionals were implicated in, and provided support for, deliberative aspects of decision-making – in particular, helping patients to access, use, and reflect on treatment or care-related information. In **Paper 3** (Discussion) I introduced the term 'infomediaries' – attributed to the Patient Information Forum (PIF) (2013). As used by the PIF, this term connoted the provision and framing of information for patients, primarily by health professionals. My synthesis revealed how not only professionals, but also family, friends, other patients (see **Papers 1**, **2**, **5** and **6**), and even researchers (see **Paper 3**) could function as infomediaries, and, perform a considerably more diverse role. My synthesis also, however, highlighted that whilst generally valued, the involvement of infomediaries could introduce tensions, and, moreover, that access to this sort of support should not be assumed.

The role and importance of infomediaries is prominent in the findings of all three studies, though my synthesis uncovered some interesting points of difference. For example, I described in **Paper 1** how relatively 'established' TYA patients reported having identified a range of people as potential infomediaries, and, in some instances, actively sought these people out. They included not only close family (in particular, mothers), but also friends or family with their own experiences of chronic illness, and wider family members with relevant professional expertise ('in-house experts'). I reported how a young man offered an example of the latter:

> *'My cousin, she's a nurse …. She'd actually written down some names of drugs to suggest … So she's had a bit of an influence too'* (**Paper 1**, p1297)

I documented further in **Paper 2** how, in making decisions about treatment of inflammatory arthritis, TYA drew on information from these wider infomediaries. I noted how TYA portrayed information provided by health professionals as important to their deliberations, but made it clear that such 'official' information was supplemented with, and reviewed against, information from other sources, including family and friends.

In contrast, TYA interviewed for study 3 identified a far more limited cast of largely self-appointed, and less influential, infomediaries: their parents (see **Paper 5**). Their parents described themselves as seeking information within and outwith

consultations – asking questions on their children's behalf, and doing research on disease, treatments, trials, and clinicians. This included turning to their own social networks for information and advice:

> *'We did have a friend who's an oncologist… She'd gone away and done her own research, came back and said, "It is a good trial… it's probably a good one to go on" – if she'd come back and said something different, we might have tried to talk (son) out of it'* (**Paper 5**, p8)

Notably, however, though these parents described feeding information back to their children, their influence on TYA(s)' deliberations and treatment decision-making appeared (from both parents' and TYA(s)' accounts) to be relatively modest. Parents often explained how they had expected and sought a relatively directive role in deliberations, particularly at diagnosis, but their aspirations were sometimes frustrated by the organisation of care. They noted how, for example, where TYA were in-patients, the provision and discussion of information by clinicians was difficult to predict. Hence, they were not party to all important conversations. TYA meanwhile appeared to privilege expert (professional) opinion over that of their parents.

With prior research (detailed in **Paper 6**) having shown that unmet information needs may result in distress and compromise quality-of-life, the role played by infomediaries (as investigators, processors, providers and retainers of information) may have ongoing value. However, it was clear from my synthesis that such individuals should not be considered neutral parties. Parents themselves often recognised that their information needs differed in important ways their children's. For example, parents reported wanting prognostic information, which might present a threat to TYA(s)' positive, recovery-focussed outlook (**Paper 5**). Other ways my synthesis suggested the role of infomediaries might be problematic included the potential for accidental or deliberate distortion of information (in order, perhaps, to 'nudge' patients towards an infomediary's favoured decision (**Paper 5**)). Confusion would also appear a potential risk, should information received through lay infomediaries not align with that provided by professionals (see **Paper 2**).

Finally, my synthesis offered a reminder that the availability of such support for decision-making should not be assumed. This consideration was initially raised in **Paper 1**. Later, in **Paper 6**, having noted how some TYA with cancer ended up

attending pivotal consultations alone – including those where their diagnosis was disclosed – I observed that patients might need prompting and/or priming to marshal infomediary support. I also reported in **Paper 5** how some TYA patients were unable to involve their parents in consultations (or indeed our research) for practical reasons, including geography and/or language. Though that did not necessarily mean that their parents could/did not take on any aspects of the role of infomediary, their involvement appeared constrained.

To recap, my synthesis highlighted the extensive informational work often undertaken in support of patients by their family and/or friends, either at the behest of patients or on others' initiative. It also indicated, however, that though generally valued, the involvement of infomediaries could in some instances be problematic.

Practical support for decision-making: Making decisions possible

Additionally, my synthesis highlighted the deeply contingent nature of decision-making, in the sense of patients being able to make decisions they believed could be implemented. Fundamentally, treatment options may only be viable for (and/or attractive to) patients if mundane practical considerations can be addressed. It was clear from my synthesis that trusted others such as close family members played an important role here – especially (though not exclusively) when aspects of the treatment(s) about which decisions were being made had to be administered outside the clinical setting (i.e., at home, school and/or in the work place).

For example, study 1 revealed how TYA with inflammatory arthritis contemplating biological therapy needed to consider numerous practicalities. These included receipt of drug deliveries, storage of medications (e.g., in a secure fridge), and their own – or others' – capacity to prepare and administer injections. For some TYA, work or study schedules and/or shared accommodation created potential challenges. Others lacked confidence in handling and self-injecting medications. In **Paper 1** I reported the work undertaken by trusted others to find workarounds for these sorts of issues, and to make decisions to start biologics appear (and be) possible. I described, in some detail, how older family members (typically mothers) often took responsibility for procuring medications, and in some instances administered these (e.g., gave sub-cutaneous injections):

> *'I order (the Etanercept)… So in a way I still have a bit of a role, that I'm checking that the injections are going down, that they're disappearing from the fridge… (and) making sure that, sort of… he's got everything he needs'*
> (**Paper 1**, p1297)

Study 3, which explored decision-making about treatment for cancer, offered more modest but still important examples of trusted others providing decision-relevant practical support. Patients in that study were often offered treatment (chemotherapy, surgery, and/or radiotherapy) administered/undergone predominantly within a hospital setting. As I noted in **Papers 5**, **6** and **7**, cancer care in Scotland is provided by 20 hospitals in 14 health boards, but certain treatments, specialist (TYA) services, and access to clinical trials, are only available at selected centres. Hence, patients electing to be treated at such centres may need to be prepared to travel some distance (frequently, and over a long period of time). For many patients in study 3, parents were a key source of transport. They enabled TYA to opt for treatment at a more distant centre (or made that a more attractive option than it would have been if TYA were dependent on hospital transport). Though a somewhat taken-for-granted form of practical support, health professionals' accounts suggested access to transport could be an important factor in decision-making about place of care and potentially trial participation (**Paper 7**).

Social considerations as a more ambiguous source of influence

My synthesis further indicated how social considerations (e.g., what people perceived others as expecting from them, and, moreover, themselves expected of life) could inform concerns and priorities in/for decision-making. It suggested such influences were somewhat ambiguous in nature and could at times be problematic, for instance diverting attention from other important considerations.

Findings from study 1, in particular, highlighted the powerful influence social considerations exerted on some patients' decision-making. TYA in that study revealed a strong desire to fit in socially, and corresponding concern with how they were perceived and judged by their peer group. TYA(s)' accounts pointed out a range of ways not only disease but also its treatment could have social costs, causing them to stand out and/or to miss out (e.g., on valued experiences). These impacts informed their perspectives on treatment. For instance, some TYA had had (and others anticipated) negative experiences of disclosure (i.e., of sharing details of

their disease and treatment) (**Paper 1**); they therefore wanted to be able to manage their condition discreetly. Others reflected on the side effects of (some) medications on their appearance – steroids, for example, causing changes to face shape, and weight gain, both of which could make them look very different to their peers.

A few TYA described how other side effects had caused absences from school, with these leading to academic underperformance, social isolation and/or bullying. TYA further observed how the need to consider medication schedules before committing to social activities had led them to miss out and/or be left out of others' plans. As I reported in **Paper 2**, these TYA aspired 'to live a normal life', a key feature of which was engaging in social and vocational activities typical of their life-stage and communities. As one health professional interviewee explained:

> *'They want to be able to get up in the morning and just be able to move. They want to go to work. They want to stay in college. They want to complete their university degree. They want to travel. They want to do normal things'* (**Paper 2**, p1291)

Hence, these young patients wanted treatments that supported, rather than challenged, these goals. I explained in **Paper 2** how this meant they favoured regimens that not only reduced symptom burden and promoted bodily function, but, in addition, were relatively simple and entailed minimal restrictions. Whilst recognising the validity of such preferences, parents and professionals expressed concerns that TYA(s)' focus on short-term priorities might be at the expense of other important considerations, in particular the longer-term effects and effectiveness of treatment:

> *You're giving [treatment] to them to help them live a normal life. But there's much more to it than that. You give it to them … to stop things happening that they couldn't even begin to imagine'* (health professional, **Paper 2**, p1292)

These TYA differed notably in their concerns and priorities from their peers participating in study 3, who were, in contrast, focused primarily on survival and recovery from cancer (see **Paper 5**). In the immediate aftermath of a cancer diagnosis, maintaining a 'normal' life was a fairly low priority. It appeared to have limited effect on these TYA(s)' treatment decision-making, though the desire to maintain an appearance and sense of normality influenced some decisions about (or at least ambitions for) self-care (see **Paper 6**).

Fundamentally, my synthesis showed how (TYA) patients wanted to be seen as normal and to live normally. Where they perceived they had scope to do so (e.g., where life was not perceived as at risk, or choices were not expected to influence survival) they made decisions supportive of such ambitions. The accounts of others (e.g., parents and professionals) raise the possibility that a focus on short-term concerns might sometimes be at the expense of important longer-term considerations.

Analytic theme 3: Service context – creating a menu and climate for decisions

My synthesis also drew attention to the (health) service context in which decision-making took place, with findings from study 3, in particular, illuminating how this could enable or constrain patients' decision-making. Three sub-themes were prominent: how policy, resource and organisational factors influenced access to treatment options; how health professionals set consultation agendas, and thereby determined opportunities for decision-making; and the overwhelming nature of clinical activity as a backdrop to decision-making. I discuss each of these, in turn, below.

Policy, resource and organisational factors influencing access to treatment options

My synthesis pointed to a number of ways in which wider (macro and meso) factors could, individually or in combination, influence access to treatment or care options, affecting (indeed determining) the decisions patients were invited and able to make. These factors included public policy and resource considerations, as well as how services and activities were prioritised and organised locally.

To illustrate, when I undertook study 1, access to certain, costly treatments for inflammatory arthritis, including biologic therapies, was (as noted in **Paper 2**) subject to patients meeting nationally agreed upon criteria. This said, in collecting data for that study, it became clear that there was considerable variation between study sites (NHS Hospital Trusts) in how stringently those criteria were interpreted and applied – and whether therefore patients were invited to make decisions about such treatments or not. In subsequent work (study 3) I explored, more deliberately, how structural factors and choices made 'upstream' of the consulting room could function to determine (limit) the decisions offered to patients. As study 3 concerned decision-making about trial participation, choices made by sponsors, investigators, and

clinical teams were all of interest. I reported in **Paper 4** how professionals' accounts suggested that, in establishing trials of treatments for rare malignancies, investigators might prioritise the involvement of larger treatment centres, in order to contain the costs of trial activation, set-up and maintenance. This limited opportunities for patients at smaller centres to make decisions about trial participation, or made these conditional on their willingness to transfer to larger centres. Principally, however, **Paper 4** focused on local practices and choices, in particular the 'discretionary' behaviours of clinical teams and individual professionals with regard to supporting and promoting particular trials. I documented how these were shaped by the prevailing system of (financial) rewards and/or sanctions, as well as competing pressures (e.g., clinical workloads, staff shortages). Notably, interviewees explained how research funding systems encouraged the opening of trials to which recruitment was likely to be straightforward, and discouraged involvement in those where it was less certain:

> *'Not to beat around the bush, we get very little money until we recruit patients. You know most of the incentivisation… bean-counting, is related to the number of recruits, not to the numbers of… studies. And so … where we may get nothing back, (our resources) would frankly be better directed to something (else)'* (**Paper 4**, p8)

Interviewees rationalized such choices as pragmatic and prudent, but their potential to affect the treatment/care options available to patients (in particular, access to cancer treatment through a trial) was clear. Interviewees themselves recognised that the prevailing logic privileged trials relevant to more common diseases (where there was a large pool of potential recruits) and functioned to curtail trial opportunities for patients (including TYA) with rarer conditions:

> *'there are some trials for very rare cancers… quite a few that are relevant to this group (TYA), where we had had to just made the decision, we don't have enough resources to open this trial that we may recruit one, or zero patients (to) over the lifetime of the trial… that's a big problem… for this group of patients'* (**Paper 4**, p9)

In a subsequent publication, **Paper 7**, I reported professionals' views on whether and how the organisation and administration of research (and care) had historically, and might in future, affect opportunities for patients in under-served groups to make decisions about taking part in a clinical trial. They surmised that there was the

potential to improve the situation (*'if we think about it… we could organise ourselves better'* (**Paper 7**, p4)) and suggested a range of ways research and care might be re-configured to facilitate greater and more equal access to trial opportunities. Their proposals coalesced around four themes: consolidating the pool of eligible patients (through centralisation of care and/or research, or more sophisticated collaboration); streamlining bureaucratic requirements (reducing duplication of activity, increasing standardisation, and establishing 'fast-track' processes for rare disease research); promoting pragmatism in trial design; and, investment in research and the research workforce.

Though these findings might seem somewhat removed from patients' decision-making, they introduce an important point. Specifically, they demonstrate that only by appreciating what goes on 'backstage' can we really understand patients' decision-making experiences. Fundamentally, if there are fewer options on the table, patients have fewer opportunities to make decisions. Furthermore, if the options are highly caveated – e.g., treatment through a trial is an option only if patients are willing to transfer to a (more) distant centre – then the choices offered to them are potentially less meaningful.

Health professionals setting the consultation agenda

My synthesis also revealed how health professionals' practices could determine patients' decisional opportunities in more direct though still subtle ways. For instance, it drew my attention to matters of how consultation agendas were set, and, more particularly, by whom.

An important finding from study 2 was that prior to the research, participating patients had been largely unaware of the connection between rheumatic disease and fatigue. **Paper 3** reported how they had consequently felt unable to raise the problem in consultations with their rheumatology team. Many of my interviewees were surprised to learn there were recognised ways of managing fatigue: none recalled ever having received any relevant professional advice or direction. Indeed, some described the initial research interview as the first time they had talked about their experiences of fatigue at any length. Contemplating possibilities for practice change, these patients said they were keen for fatigue to have a place on consultation agendas, with health professionals initiating discussions about its management:

> *'It would be great if the consultants did say to you, "And how are your fatigue levels?" But that doesn't happen. It doesn't happen'* (**Paper 3**, p233)

Such experiences point to the critical role health professionals play in setting consultation agendas, and legitimising patients' experiences and concerns, as a precursor to presenting and discussing options (whether medical or self-care regimens) for the management of troubling symptoms.

Additionally, findings from study 1 revealed how, in the absence of opportunities to air their concerns, patients might seek their own solutions. I described in **Paper 2** how, lacking an explicit invitation to share difficulties associated with the physical or psycho-social side effects of a treatment, TYA patients did not necessarily disclose these. As a result, alternative treatment options were not discussed. Some then made (arguably sub-optimal) unilateral decisions about discontinuing or modifying their treatment regimen. Care teams only become aware of these decisions when marked deterioration occurred, or some other sort of crisis point was reached. A key conclusion I drew from those findings was that, in order for sustainable treatment plans to be established, TYA needed (early and regular) opportunities, and moreover active encouragement from health professionals, to share their treatment experiences, concerns, and difficulties. In essence, space needed to be made on consultation agendas for patients' concerns and priorities to be aired.

The nature of clinical activity: complex and overwhelming

Finally, my synthesis highlighted how the nature of clinical activity functioned as a backdrop to, and created a particular (often challenging) climate for, patients' decision-making. Findings from study 3 illustrated especially vividly how the unfamiliarity, complexity and relentlessness of clinical activity could overwhelm patients, presenting further challenges to deliberation and decision-making.

To explain, I reported in **Paper 6** how TYA with cancer had often had a relatively poor understanding of the purpose and implications of their tests, referrals and care pathways in the lead-up to diagnosis. They were consequently ill-prepared for the cascade of decisions with which they were then confronted. For example, I documented how, in the absence of any real appreciation of their diagnostic trajectory, some TYA were bewildered to find the hospital to which they had been sent was a specialist cancer centre:

> *'I was a bit confused. I didn't really know what was up with me. And then obviously at the (Cancer Centre) it says (Regional) Cancer Centre, whatever it is, underneath the (Hospital Name). So I was like, "Right, what's, what's going on?"'* (**Paper 6**, p6)

Understandably such TYA were discombobulated and ill-prepared for the conversations that then took place, making it harder to marshal their concerns, ensure these had a place on the consultation agenda, and negotiate relevant information and decisional opportunities.

Moreover, many TYA in that study described how diagnosis, when it came, served to trigger a rapid escalation in clinical activity, beginning (where diagnosis was imparted by a health professional other than an oncologist, e.g., the surgeon who had undertaken the decisive biopsy) with onward referral to new teams, departments and often new hospitals. In **Paper 5** I documented how TYA (and caregivers) recalled 'a whirlwind of activity', leaving little time to process and reflect on their diagnosis and any information on treatment given to them. TYA described undergoing an assortment of tests and procedures in the aftermath of diagnosis, the purpose of which was to confirm (the specific type of) cancer, and/or establish its spread, as a basis for treatment planning:

> *'after I saw my (Consultant Oncologist) for the first time, he arranged loads of different scans. Because it was very common for it to, have spread elsewhere in the body, particularly the lungs, and possibly the bones. Obviously that was extremely harrowing. I got a CT scan, I got a bone scan, I got a full body scan, heart scan, kidney scan'* (**Paper 5**, pp5-6)

In addition, several interviewees described having had consultations about fertility, and procedures to try and preserve this, between diagnosis and treatment initiation. Some TYA, such as the following young man, welcomed the forward momentum of care:

> *'it's just been literally full steam ahead … and I've preferred it like that because you've not got any time to think about things'* (**Paper 5**, p6)

However, others highlighted the challenges this created for reflection. With time and energies rapidly eroded by the battery of clinical interventions set in train, the tempo of activity was far from conducive to informed, and deliberative, decision-making.

Closing comments

To summarise these findings, my synthesis highlighted the profound importance of the personal, social, and service contexts of decision-making. Drawing on seven publications, I distilled out a series of relevant themes and described how: (1) in different ways, experiences of physical, emotional, and cognitive dis-ease shaped and informed patients' decision-making; (2) family and friends acted as enablers (but social considerations influenced decision-making in more ambiguous ways); and, (3) policy, resource, and organisational factors, and the discretionary behaviour of individual clinicians, acted to determine the choices patients were given, whilst clinical activities sometimes created conditions at odds with deliberative decision-making. Next, I consider how my findings relate to the work of others – more specifically, how they fit with, support and/or challenge the wider literature, both SDM scholarship pre-dating my work, and more recent relevant literature from within and outwith that field.

REVIEW OF MY FINDINGS: THEIR RELATIONSHIP TO WIDER SCHOLARSHIP

My synthesis has produced a new set of 'findings', inviting a more complex understanding of the factors at work in patients' health-related decision-making. In outline these findings take the form of an overarching proposition, and a series of analytic themes (underpinned by descriptive sub-themes) that, drawing on Glaser (1965: 437), are both 'plausible and close to the data'. My proposition is that ***health-related decision-making happens in and is shaped by context***. The analytic themes are: *personal context – experiencing a state of dis-ease*; s*ocial context – a source of support and influence;* and *service context – creating a menu and climate for decision-making.* I now consider how my findings align with, challenge and/or complement wider scholarship. Specifically, reflecting upon each theme and sub-theme in turn, I:

- revisit the theoretical and empirical work on SDM pre-dating the submitted publications (discussed in more detail in the Introduction);
- consider how my findings relate to more recent SDM literature; and
- reflect on parallels and/or potential synergies with literature from *other* fields/disciplines (including medical sociology, and various branches of psychology). This approach is consistent with wider practice in applied health services research, which finds value in literature according to its relevance, rather than its disciplinary origins.

I close this section of the critical review by reflecting on the strengths and limitations of (both) the contributing publications and the process of synthesis/review, before considering the implications of my work for further research and practice.

Analytic theme 1: Personal context – experiencing a state of dis-ease

My finding that patients were often experiencing physical, emotional and/or cognitive dis-ease when invited to make decisions about their treatment and/or care – and, moreover, that these dis-ease states could influence decision-making in powerful ways – may appear somewhat self-evident. Yet the classical SDM literature has paid only cursory attention to the presence and potential implications of these different forms of dis-ease. I discuss this further below, noting how support for my claims can be found in more peripheral SDM literature (e.g., that authored by medical

sociologists) and in research emerging from other disciplines, in particular psychology.

Physical dis-ease and decision-making

My synthesis highlighted the ubiquitousness of physical dis-ease, with pain and fatigue – associated primarily with symptoms but also arising from clinical interventions – being common manifestations. It revealed how these (and other) forms of physical dis-ease could affect decision-making via, *inter alia*, their impact on patients' capacity (to engage with, process, and use decision-relevant information) and access to resources, including experiential knowledge.

Early SDM literature, in contrast, gave limited attention to physical dis-ease – beyond acknowledging that interest in being involved in decision-making might be affected by the type and severity of patients' illnesses, and, in the most acute medical situations, their physical condition might preclude involvement (The Health Foundation, 2012). There are some notable exceptions, such as Rapley (2008:440), who argued that more attention should be given to the importance of 'bodies, emotion… and suffering'. Rapley is a medical sociologist and his position echoes the argument made by colleagues including Lawton (2003) that the physical and other 'mundane' aspects of illness warrant closer scrutiny when seeking to understand patients' experiences. It was also no doubt informed by the seminal literature of the discipline, which has illuminated how the nature of people's symptoms and treatment (side) effects shape their personal illness experiences (Charmaz, 1983; Bury; 1991).

Recent years have seen the emergence of a small body of literature on pain and SDM. However, that work attends almost exclusively to SDM <u>about</u> pain management, rather than the implications of pain <u>for</u> SDM (see, for example, Holland et al. (2016), Wegier et al. (2020) and Matthias and Henry (2022)). For insights on the implications of pain, it is necessary to look to other fields/disciplines. Literature emerging from (experimental) psychology, neuroscience, and anaesthesia, offers findings of relevance and interest. For instance, evidence (covering diverse populations) suggests pain may affect decision-making (generally) by diminishing cognitive functions such as attention, memory, mental flexibility, and reasoning (Baliki et al., 2008; Moriarty & McGuire, 2011; Aja, 2017; Cowen et al., 2018; Salcido et al., 2018; Barnhart et al., 2019; Khera & Rangasamy, 2021; Phelps et al., 2021).

Moreover, chronic pain may induce enduring changes in the brain; in particular, in regions associated with higher order executive functions. As yet, the implications for health-related decision-making remain to be fully understood. However, such evidence provides broad support for my finding that physical dis-ease, in the form of pain, may introduce challenges with regard to people absorbing, processing and employing critical information about their diagnosis and treatment options.

To date, fatigue has received yet less attention in the SDM literature than pain, with even literature regarding SDM about fatigue management being hard to locate. This is in some respects surprising, as although invisible, non-specific and still poorly understood, fatigue is now widely recognised as common to much chronic illness (Menzies et al., 2021). A notable exception is the work of Teshale et al. (2019), which explored associations between people's scope to make independent decisions, fatigue, and quality of life. Interestingly, their findings suggested that increased decisional autonomy might improve both management of fatigue and overall well-being. Again the effects of fatigue on decision-making appear to remain under-researched, though Menzies et al. (2021) noted evidence suggesting that the enduring fatigue associated with chronic illness may cause cognitive impairment, which (by inference) might create challenges for health-related decision-making consistent with those reported in my synthesis.

My synthesis additionally highlighted how experiential knowledge ensuing from prior episodes of physical dis-ease functioned and was drawn upon as a resource in decision-making. Once again support for this finding can be found in the writings of medical sociologists, and, additionally, those of (health) psychologists. For example, Ziebland & Herxheimer (2008) similarly described how, when making health-related decisions, people drew on and were influenced by evidence derived from personal experience. This finding was echoed in recent research on young people's cancer-related decision-making (Darabos et al., 2021). Notably, my synthesis documented how (recourse to) such experiential knowledge could both influence people's priorities and concerns with regard to future treatment and care options, and affect the character of their decision-making. In particular, I noted how awareness of the uncertainties of treatment appeared (outwith acute disease scenarios) to foster more experimental, and provisional, approaches to decision-making. This finding too

derives support from work arising from medical sociology: Conrad (1985), Bury (1991), Donovan & Blake (1992), and Pound (2005) all portrayed patients as active agents, accruing knowledge over time and undertaking careful treatment 'experiments', progressively implementing, assessing and modifying their treatment decisions.

Emotional dis-ease and decision-making

My synthesis also drew attention to the widespread presence of emotional dis-ease, noting how patients (and other research participants) recalled experiencing deeply uncomfortable and/or challenging emotions whilst facing important treatment decisions. This finding is consistent with Ferrer & Mendes' (2018:1) assertion that: 'health decisions … often take place in emotionally-laden contexts'. Yet within the SDM literature, decision-making has historically been conceived of – and researched – as a rational cognitive endeavour, with efforts to understand health-related decision-making consequently focussed on a narrow set of determinants, principally information and knowledge (Ferrer & Mendes, 2018). In this, the SDM literature largely parallels that of other disciplines with interests in decision-making (e.g., behavioural economics and psychology) where, for most of the 20th century, rational choice models dominated work, with the role of emotion receiving minimal attention (Lerner et al., 2015).

In psychology, however, there has been a shift in recent years, with increasing recognition that emotions may play a role in decision-making – and interest in seeking to understand exactly how. Emerging work is relevant to, though rarely focused on, health-related decision-making. Research in the new sub-discipline of emotion science has explored the effects of both 'integral' emotion (emanating from a particular situation) and 'incidental' emotions (unrelated emotion, carried over/into one situation from another) with some scholars concluding:

> *'emotions constitute potent, pervasive and predictable, sometimes harmful and sometimes beneficial drivers of decision making.'* (Lerner et al., 2015:799)

Others (e.g., Luo & Yu, 2015) have asserted that the time has come for the focus of empirical research to shift from whether emotions affect decision-making, to when and how this happens. Such work, they have argued, should include attending to how emotion interacts with reason to shape the decisions people make. My own

findings strongly suggest that such interaction happens, illustrating how emotion (emotional dis-ease) may act alongside, and in conjunction with, cognitive activity and dis-ease to impede engagement with, absorption and processing of important information on disease and treatment (thereby undermining decision-making capacity). This resonates with findings from psychology – that emotion can affect attention, cognitive processing, how problems are framed, orientation to risk, and observance of rational action (Lerner et al., 2015; Kozlowski et al., 2017).

It has additionally been suggested that different emotions (such as fear and anger) may have varying effects, and, moreover, whilst potentially presenting challenges to decision-making, emotion may also in some instances facilitate decision-making (helping people reach decisions which are congruent with their values) (Hermann et al., 2016; Carpenter & Niedenthal, 2018; Chick, 2019). Chick (2019), for example, drew attention to mechanisms including: emotion as a source of unique – and sometimes beneficial – information; emotion as a tool for the efficient processing of high volumes of information; and emotion prompting the privileging of particular, affectively salient information. Hence, whether and how emotion ultimately impedes or facilitates decision-making might depend on the particular emotion, the nature of any given decision, and other characteristics of the decision-maker. Thus, many of the authors cited above have called for more detailed, empirical work, drawing on patient narratives and contextual information, to understand how different emotions influence different patients, and are managed and used in the making of different decisions (Hermann et al., 2016; Carpenter & Niedenthal, 2018).

Though a body of incidental findings is mounting, as yet research attending (explicitly) to emotion, mood and health-related decision-making remains exceedingly limited – a gap Ferrer & Mendes (2018) cited as important, and worthy of being filled. Ferrer & Mendes (2018) have additionally called for greater ‘crosstalk’ between disciplines with cognate interests, noting that similar research concerns are framed differently depending on the disciplinary lens and asserting that, as yet, there has been a failure to integrate perspectives and knowledge effectively. Certainly the emerging literatures reported here appear to have had limited impact on/within the field of SDM, though some recent review papers offer tentative indicators of change (Mazzocco et al, 2019; Treffers & Putora, 2020).

Cognitive dis-ease and decision-making

The third, and final type of dis-ease highlighted by my synthesis is cognitive dis-ease. Closely intertwined with the forms of dis-ease discussed above, my synthesis revealed how cognitive dis-ease could manifest in different ways, including as uncertainty and/or (a sense of) overload. I noted how patients' responses included selective attendance to information, with this having potentially problematic consequences for decision-making.

Beginning with uncertainty, I described how this appeared, typically, to arise from lack of knowledge and/or ambiguity of information. This finding is consistent with the work of Alquist et al. (2020), who proposed that uncertainty involved people lacking information, or being unable to judge the correctness of the information available to them. Gulbrandsen et al. (2016) offered a subtly different definition, describing uncertainty as arising from the *perception* of ignorance. Uncertainty as construed by Alquist et al. (2020) is perhaps easier to resolve, as, logically, lack of information might be remedied through provision of information. Indeed, this is a central plank of SDM (which, as noted above, conceives decision-making as a rational, cognitive activity, in which information plays a central role (Ferrer & Mendes, 2018)). However, recent writings such as that of Fisher et al. (2018) have started to problematize the central assumption of SDM that if an appropriate (SDM) protocol is followed, and available tools such as decision aids used, patients can, in general, be transformed into informed and knowledgeable decision-makers. Fisher et al. (2018) argued that in many instances (i.e., clinical scenarios) the existing evidence is not only complex, but also incomplete, ambiguous, and even conflicting. Providing patients with information about the evidence base, as part of the process of SDM, might therefore, they mooted, actually function to heighten uncertainty, by making patients more conscious of, and sensitive to, both deficiencies in their own knowledge and the limitations of wider understanding.

Nevertheless, the imperative to *try* to inform patients fully seems strong. But is it also possible to have too much information, pertaining to too many options? The findings of my synthesis – which highlighted patients' experiences of being informationally (and cognitively) overwhelmed – suggest the former certainly is true. A number of other scholars (working across a range of areas/disciplines) have made similar

assertions. Peters et al. (2013) noted that whilst the assumption underpinning much public policy was that more information, about more options, would produce better outcomes, psychological research indicated that more information, options, and freedom (to choose) could also have negative effects. Informational overload, in particular, could sometimes lead to cognitive overwhelming and poorer decisions. Peters et al. (2013) further noted that whilst decision aids may be intended to help people identify what is important when confronted with large volumes of information, in practice many exceed patients' abilities to understand and use them. Bester et al. (2016) also considered how informational and cognitive overload (due to either the volume or complexity of information) could overwhelm decision-making capacity and compromise informed consent. However, they surmised that the point at which overwhelming might occur would depend on both the nature of the information and the character of individual patients. Discussing the implications of this for SDM (and vice versa) they suggested that efforts to inform patients about their options and the implications of each course might be more beneficial where emotional overload was the issue, than where cognitive capacity was already stretched.

Returning to my own work (my synthesis) I discussed how cognitive (and emotional) dis-ease might trigger potentially problematic behaviours such as selective attendance to information and/or avoidance of that which was cognitively (or emotionally) challenging. Whilst scanning the available information and deciding what is relevant is a normal process, it is important that people give attention to the right things. Peters et al. (2013) noted how cognitive and emotional stresses could result in people giving undue attention to certain pieces of information and allowing them disproportionate weight in decision-making. The work of others offers some insights into why and how this might happen. Alquist et al. (2020) described findings from psychological research suggesting that situational uncertainty prompted efforts to conserve energy (for an uncertain future). These led to diminished performance on tasks involving executive function (higher order cognitive processes). They surmised that in the context of decision-making, such a shift to low-energy/effort responses might involve focusing on an isolated aspect of a decision/option, and failing to take the care needed to prevent logically irrelevant information from biasing decisions. This might plausibly result in sub-optimal decisions.

To sum up, in attending to dis-ease, my work helps to fill important gaps in the decision-making literature. Echoing and extending research undertaken by medical sociologists working within the field, my synthesis highlights the importance to (and potential influence on) decision-making of three common forms of dis-ease. Interestingly, work in other research fields and social science disciplines, in particular psychology, though appearing to have had limited influence on the trajectory of SDM research, supports and complements my findings. That wider work, like my own synthesis, suggests that physical, emotional, and cognitive dis-ease may have profound effects – and, moreover, that the relationship *between* them is complex and dynamic (mutually influencing and reinforcing).

Analytic theme 2: Social context – a source of support and influence

Next, my synthesis highlighted the importance of social context. I drew together material illuminating how family, friends and others provided deliberative and/or practical support, thereby acting as enablers of decision-making. Alongside this I observed the potential for social considerations to have more ambiguous (less unequivocally positive) effects. There is a clear tension between the first of these findings and the classic literature and models of SDM; there are, however, parallels in/with work informed by medical sociology and more recent research emerging from the field of SDM.

Deliberative support: the role and importance of 'infomediaries'

My synthesis highlighted and emphasised the important informational work family and/or friends undertook in support of decision-making. To capture this, I introduced the term 'infomediary', explaining how such a role might involve a range of activities and be taken on at the behest of patients, or self-assumed (i.e., unilaterally adopted). I noted that, though generally valued, the involvement and work of infomediaries varied in its influence and could, at times, be problematic.

In attributing such an important role to others, my synthesis contrasts with the classic literature and models of SDM, which largely conceived decision-making as something that involved a discrete encounter between two parties (a clinician and a patient). Such models were informed by the ethical principle of autonomy (Beauchamp & Childress, 1979) – and more particularly a rational and atomised version of autonomy. However, early sociologically-informed work on significant

others' involvement in decision-making (e.g., Ohlen et al., 2006; Rapley, 2008) produced findings more closely aligned with my own. Moreover, recent years have seen a further shift in perspective, with SDM scholarship drawing, increasingly, upon feminist concepts of relational autonomy (Ho, 2008; Walter & Ross, 2014; Shih et al., 2018). There is burgeoning recognition that consultations may involve and be shaped by multiple parties (e.g., several health or other professionals, familial caregivers/supporters) as well as intrusive technologies (phones, computers, and electronic patient records) (Swinglehurst et al., 2014; Bunn, 2018).

An expanding body of research documents the range of informational and decision-supportive tasks undertaken by family (and friends). For example, Asiedu et al. (2018) described how, in support of patients with ovarian cancer's decision-making about clinical trial participation, family members shared knowledge, attended appointments, and contributed to relevant discussions. They observed how patients viewed such relationships as valuable resources in decision-making. Similarly, based on a study of (women's) decision-making about the management of breast diseases, Shih et al. (2018) reported how patients depended upon family members to seek information on their behalf, take notes in consultations, synthesise information, and liaise with professionals about matters such as the practicalities of care. This, they noted, was particularly so at times of emotional upheaval, when patients felt their capacity for reasoned decision-making was diminished. Documenting the experiences of people with advanced cancer, Dionne-Odom et al. (2019) likewise described how family caregivers undertook several distinct roles in support of deliberation, including information seeking, facilitating discussions with patients about their concerns and priorities, posing 'what if' questions, and ensuring a common/shared understanding of any plans made.

I observed in my synthesis how care processes (e.g., signposting and scheduling of key conversations) did not appear to consistently support the work undertaken by family/friends. For example, I highlighted how parents of TYA with cancer expressed frustration at being unable to anticipate, and therefore take part in, some important discussions about their son/daughter's treatment and care. Dionne-Odom et al. (2019) similarly reported how family members described feeling unsupported in, and/or actively excluded from, important parts of the decision-making process. Furthermore, as their data suggested that family caregivers often played critical

roles, influencing both the process and outcomes of patients' decision-making, Dionne-Odom et al. (2019) argued convincingly that this (exclusion) was problematic, and a cause for concern.

Like Dionne-Odom et al. (2019), my recent work (study 3) explored the experiences of cancer patients and their families. Oncology has been described as a specialism where (compared to some others) caregiver involvement in consultations and decision-making is relatively widespread and accepted (Schuster et al, 2020). Nevertheless, as Laidsaar-Powell et al. (2018a) noted, communication training for oncology professionals has historically focussed on communication with the patient (only). Encouragingly, new guidelines have recently been published, intended to support and provide strategies to help professionals communicate effectively with families of cancer patients (Laidsaar-Powell et al., 2018a; Laidsaar-Powell et al., 2018b). Consistent with my own findings, these guidelines recognise, and caution, that caregivers may have emotional and informational needs which are important, but distinct from those of patients (Laidsaar-Powell, 2018a).

Other authors have highlighted the (yet more) limited interaction between caregivers and clinicians in other specialisms including in-patient mental health care, attributing this to what they view as the inadequate conceptualisation of caregiver involvement in models of SDM. Specifically, Schuster et al. (2020) have criticised the dominant frameworks for SDM for not explicitly involving – or offering clear guidance for the inclusion of – caregivers (or other 'third parties'). My own work – like theirs – supports the calls of others (Hamann & Heres, 2019; Waldron et al., 2020) for SDM models (including the revised 'Three-talk' model (Elwyn et al, 2017)) to be developed further, to explicitly incorporate family/caregiver involvement.

Practical support (how others make decisions possible)

My synthesis additionally highlighted how family (and/or friends) may contribute by providing the practical support needed for decisions to be real, meaningful (i.e., workable) options. Conventionally, SDM research and writings have focused on communication within consultations. Limited attention has been given to the conditionality of decisions, whether they are ultimately implemented, and the role trusted others play in this. As I noted earlier, the implementation of many treatment/care practices 'decided on' in clinic is contingent on resolving mundane

practical issues. Moreover, such practices are often difficult, tedious, and/or frightening (Mol, 2009) and decisions often have, in effect, to be (re-)made on a daily basis, in the context (and against the challenges) of one's life experiences or 'lifeworld' (Pickard & Rogers, 2012).

Medical sociologists, philosophers and health services researchers have all argued that patients' capacity to marshal family support can be critical to the way illness is experienced and managed. For example, Lawton (2003) stressed that to understand patients' experiences fully, it is necessary to attend to what she termed 'micro-factors', such as family and intimate relationships. This position builds on work by Bury (1982) and Charmaz (1983), both of whom wrote extensively about the ways social circumstances and resources (e.g., what Charmaz termed 'supportive intimates') influence people's experiences of illness. Offering some concrete examples, Ho (2008) noted how family members not only accompanied patients to consultations and other appointments (e.g., for investigations or procedures), but also provided deeply practical support; visiting and bringing patients food whilst they were in hospital, and providing personal and more medically-oriented care at home after discharge. Writing more recently, Doekhie et al. (2020) reported how older adults confronted with decisions valued such practical/instrumental support from close family highly (notwithstanding that it could also, at times, feel oppressive). They further observed that a perception of having familial support predicted engagement in (i.e., the sharing of) decision-making by older adults in their study.

Building on such literature, and echoing Rapley (2008)'s work on distributed decision-making, Clayman et al. (2017) contended that the early characterisation – and ongoing perpetuation of the idea – of decisions as discrete, unchangeable events that happen in (and only in) such scheduled medical encounters is a significant weakness of prevailing models of SDM. In support of this criticism, Clayman et al. (2017) highlighted evidence that many decisions 'made' in a consultation are not meaningful in the sense of ever being enacted. For example, drawing on research around adherence, they reported how around one-third of all prescriptions go unfilled, whilst in other instances medications are not taken as prescribed, and not all patients attend for follow-ups or follow-through on referrals to other services. My own work both recognises the practical and logistical conditions of many treatment decisions, and illuminates the diverse and important roles family

(and other informal carers) play when it comes to making their enactment appear and actually *be* possible.

Social considerations as a more ambiguous source of influence

My synthesis suggested that social considerations (e.g., expectations and/or perceived responsibilities) might further influence patients' decision-making, including in less unequivocally positive ways.

For instance, I reported how such considerations shaped patients' concerns and priorities when making decisions, and their broader aspirations for treatment and care. To begin with, I described how young people with arthritis aspired to fit in socially, and not to stand-out from or be identified as 'different' by their peers. Hence they were concerned about the effects of treatment on their appearance as well as if/how treatment might signal their differences. The classic literature of medical sociology would suggest such concerns are reasonable – Bury (1982) noted the precarity of social relationships, and documented the efforts people consequently made to manage appearances and sustain their social position.

These young patients also wanted 'to live a normal life' in the sense of sharing in the experiences of their peer group and made treatment decisions they considered supportive of that. Whilst it is tempting to attribute the desire to appear, feel, and live like one's peers to youth, research into the concerns and priorities of older adult patients has generated similar findings. For instance, Cornelissen et al. (2021) reported that what mattered to (older) patients with rheumatoid arthritis when making treatment decisions was (maintaining) independence, being able to meet others, and – in their own way – leading 'a normal life'.

I further noted how some parents (and professionals) were concerned that the desire to fit in, and live a normal life, could encourage a focus on short-term concerns, at the neglect of longer-term considerations. Other authors have described additional ways in which social expectations, and perceptions of what is 'normal', may prompt problematic – or at least unhelpful – decisions about help-seeking and/or care. For example, Sanders et al. (2002) described how older adults might not seek, nor therefore receive, beneficial treatment for osteoarthritis, due to perceptions of joint pain and associated disability as a 'normal' and inevitable part of ageing.

The work of Bury (1991) and others suggests further ways in which social considerations may influence decision-making. One of these is the impact patients perceive their treatment choices as having on others, in particular their family. This impact can take many different forms, including financial (e.g., loss of earnings, direct costs of treatment) as the work of Exley et al. (2012) illustrates. Their research on individuals' decision-making about paying for high cost dental treatments (implants) showed such decisions to be mediated by a range of factors, including the impact the expenditure was expected to have on family members.

Some scholars have questioned whether attention to family interests could (potentially) compromise patient autonomy. For example, Blackler (2016), a biomedical ethicist, reflected on the potential for discordant expectations to emerge between (cancer) patients and their families, leading to tensions with regards to decisions about treatment and, in particular, end-of-life care. Blackler suggested such misalignments could generate concerns within the health/care team that patient autonomy might be compromised. Others, whilst acknowledging such concerns, have stressed that compromised autonomy is not inevitable. Some such as Ho (2008) have gone further and suggested that family involvement and attention to family interests may actually promote patient agency. This latter argument is consistent with more interdependent, 'selves-in-relation' to others (Hallowell, 1999:616), or relational concepts of autonomy, which over recent years have begun to hold increasing sway.

To re-cap, my core finding that family and friends (often) provide important forms of support for decision-making challenges the classic literature and models of SDM, which have favoured, and functioned to promote, the principle of individual autonomy. It is, however, consistent with work informed by the traditions of medical sociology. Moreover, it complements and reinforces critical perspectives emerging from within SDM, such as that of Clayman et al. (2017), who highlighted the need to understand the complicated realities of decision-making, including the influence of family within and outwith medical encounters. Whilst demonstrating that the involvement/influence of others may be very valuable, my work also recognises that it is not unequivocally positive. Fundamentally, my synthesis has shown that social context may support – but also sometimes challenge – the promotion of high quality

decision-making. It seems both important and possible that models of SDM begin to acknowledge and account for this.

Analytic theme 3: Service context – creating a menu and climate for decisions

Finally, my synthesis illuminated how policy, resource, and organisational factors, as well as the decisions and behaviours of individual clinicians, influenced the options available to patients, and the decisions they might therefore make. It also illustrated how clinical activity functioned as a backdrop to, and created an often challenging climate for, patients' decision-making. Though early SDM scholarship provides limited support for (or, conversely, challenge to) these findings, other literatures and recent developments in the field (of SDM) suggest an emergent recognition of, and interest in, service, organisational, and structural influences.

Policy, resource and organisational factors

Twenty years ago, Wildes (2001) – a bioethicist – noted how clinical encounters were framed and shaped by the financial, regulatory and administrative structures of healthcare delivery, medical education and research, and Lawton (2003) – a medical sociologist – emphasised the importance of locating patients' experiences in wider, collective contexts. However, coeval SDM scholarship gave little consideration to such system or service-level factors. Outlining a multi-disciplinary model for SDM a decade on, Legare et al. (2011) acknowledged the *potential* for health system-level factors to influence activity within clinical encounters. Nevertheless, they did not delineate what those factors might entail.

My own research, and synthesis, has drawn out some very concrete ways in which regulatory, financial, and administrative forces may affect both patients' opportunities for, and experiences of, decision-making about treatment and care. In particular, I have documented how policies and systems of financial reward (and sanction) for involvement in research activity may influence local decision-making about whether and which clinical trials to open – and hence patients' opportunities to make decisions about receiving treatment through a trial. This advances understanding, firstly, of the complex and varied influences upon health-related decision-making, and secondly, of access and recruitment to trials specifically (where my work complements literature detailing how issues of individual equipoise and role conflict

may affect recruitment practices (Garcia et al., 2004; Donovan et al., 2014a; Donovan et al., 2014b; Guillemin et al., 2017)).

While my own findings relate predominantly to structural influences on opportunities for decision-making about trial participation, other contemporary research has documented the influence of regulatory and financial factors on quite different sorts of health decisions. For example, McDonald et al. (2012) reported how dentists in England described altering their practices following the introduction of a new service contract. Specifically, dentists portrayed new arrangements as encouraging them to focus on achieving contractual targets, and discouraging them from offering and undertaking more complex (and costly) procedures. Another study in English dentistry (Vernazza et al., 2015) highlighted the influence of a range of non-clinical considerations, including professional codes, legal obligations and commercial factors, when dentists considered if and how to present dental implants as treatment options to/for edentulous patients.

Reflecting on the impact of (UK) funding arrangements for other areas of healthcare, Joseph-Williams et al. (2017) surmised that in some instances these too might curtail options and discourage SDM. For example, they noted how the Quality and Outcomes Framework incentivised certain (evidence-based) practices, irrespective of whether these aligned with patients' priorities, concerns or preferences. Alongside this, they observed how tensions were emerging between adherence to clinical guidelines, referral management schemes, and enactment of SDM. Offering a perspective from the United States (US), Clapp et al., (2021) highlighted how payment models often strongly incentivised surgical intervention, even where the clinical necessity of procedures was debatable. Meanwhile, Thomas et al. (2021) noted how insurance and/or reimbursement models incentivised both the use of particular treatments, and swift processing of patients, neither of which could be considered conducive to meaningful SDM. Other literature offers a counter-point, illuminating how legislative, financial and policy arrangements can conversely *promote* SDM. For example, Spatz et al. (2017:1309) reported how several US states had passed legislation encouraging SDM for elective joint replacement procedures, and Medicaid had made SDM a condition of payment for certain other interventions.

With regard to organisational influences, my own work has highlighted how workforce issues, such as workload and staff shortages, can influence both the opening of and recruitment to trials, and thereby patients' opportunities to make decisions about participation. The last few years have seen a surge in interest in the impact, on SDM, of factors including workforce planning and development, care pathways, processes, environments and practice cultures. Hence, my work speaks to an emerging body of SDM-relevant research and theory informed by realist(ic) evaluation, organisational theory, and implementation science. Examples of such work include Waldron et al. (2020), who explored the system-support, or service conditions, necessary for SDM to become a routine feature of practice. As well as resourcing for adequate staff time, Waldron et al. (2020) identified facilitators including policy, training and tools. Other research suggests a host of further organisational conditions or factors may potentially constrain or enable SDM. There is not scope to discuss these factors in depth here, but they have been said to include: physical environments (noise levels and privacy); workflow factors (scheduling, continuity of care, access to information including electronic health records, and multi-disciplinary team processes); service priorities (leadership, incentives and rewards, clinical guidelines and quality assurance tools); and, service cultures (Pilnick & Zayts, 2015; Hamilton et al., 2016; Bunn et al., 2018; Scholl et al., 2018; Waddell et al., 2021).

Health professionals setting the consultation agenda

My synthesis also drew out how professionals' consultation management practices could set the parameters for patients' decisional opportunities. In particular, it revealed how the control health professionals had (or were perceived to have) over consultation agendas meant some issues (e.g., fatigue, psycho-social impacts of treatment) and associated or alternative management options were never discussed. It called attention to consequences including: lost opportunities for patient involvement in decision-making; unaddressed symptoms with quality of life impacts; and unilateral decision-making (undisclosed non-adherence to treatment regimens).

Understanding what matters to patients has been a prominent concern of both medical sociologists and SDM scholars – albeit these groups have framed their interests in different terms. In a contribution to the sociological canon, Bury (1991) long ago noted how patients' personal priorities (goals) might differ substantially from

those of clinicians. SDM scholars (see for example, Elwyn et al., 2012) subsequently stressed the importance of exploring patients' values and preferences, alongside making them aware that different treatment options exist, and detailing what those entail. Fundamentally, however, options are responses to a problem: my synthesis suggests that who gets to define the actual problem (e.g., fatigue, psycho-social impact of treatment), and ensure consultation time is given to its discussion, is something that warrants closer attention.

Interestingly, agenda-setting featured prominently in the early patient-centred care literature. Specifically, Levenstein et al. (1986) suggested that a distinguishing feature of patient-centred practice was attention to patients' agendas as well as physicians, and a commitment to reconciling the two. Other literature broadly concurrent with my own research offers complementary and interesting insights on the topic of consultation agendas. For example, Matthias et al. (2013) published a commentary arguing that whilst much SDM work had focussed on a small component of the clinical encounter, experiences were shaped by the entirety of the interaction and the quality of relationships developed therein. Hence, they argued, eliciting patients' concerns and agreeing on an agenda together was essential to creating an environment conducive to SDM. Matthias et al. (2013:177) proposed that 'agenda setting is really the first SDM opportunity of the consultation': it both is, and enables, SDM.

Moving on in time, current policy/grey literature explicitly advocates collaborative agenda-setting. For instance, recent National Institute for Health and Care Excellence (NICE) guidance (summarised by Carmona et al., 2021) highlights the importance of co-constructing agendas, and strongly recommends agreeing an agenda for conversation as a strategy for incorporating SDM into (routine) practice. My synthesis provides support for such recommendations, notwithstanding the challenges (e.g., time) they may conceivably introduce.

The nature of clinical activity: complex and overwhelming

Finally, my synthesis highlighted how clinical activity could create a complex backdrop for decision-making, and indeed proved quite overwhelming for some patients. These findings are supported by a small but powerful body of work

emerging from service design, SDM, health services research, and implementation science.

For example, I reported how TYA patients were discombobulated by care pathways, tests undertaken, and in some instances the results received prior to their cancer diagnosis. Griffioen et al. (2021) argued persuasively that the impact of patients' experiences accumulates, functioning over time to support, or undermine, their capacity to participate in decision-making. Griffioen et al. highlighted the demands on patients of gathering, understanding and applying information, and emphasised the unpredictability, to many, of key decision points (echoing another of my findings). Consistent with my own position, they argued that 'the stage for SDM is often set outside the consultation, which might explain the limited effect currently seen of interventions focusing on consultation itself' (Griffioen et al., 2021: 5913).

I further documented how TYA with cancer described diagnosis as followed by swift onward referral (to new teams/services) and rapid escalation of clinical activity. This included further investigations, and intervention to address immediate problems or limit disease progression, whilst longer-term treatment plans were made. Reflecting on patients' experiences in the intensive care unit, Clapp et al. (2021) similarly noted how a sequence of tests and procedures would be initiated on admission, with one intervention often prompting or necessitating another. Clapp et al. likewise observed how momentum could build very quickly, with decisions subsequently being made against tumultuous clinical backdrops, which appeared to offer little room for deliberation by patients and/or families. I explained in my synthesis how escalation in clinical activity imparted a sense of urgency, discouraging patients from taking time for reflection. Writing about older women's experiences of decision-making about participation in ovarian cancer trials, Asiedu et al. (2018) similarly observed how these were often characterised as pressurised. They noted that notwithstanding consent dialogue and documents emphasising the importance of taking time to deliberate, trial design factors and/or the organisation of care appeared to work against this. Some scholars have questioned whether it is ultimately ethical to ask patients to make decisions about trial participation in pressured scenarios including acute medical situations and neonatal emergencies (Fern et al., 2008; Buckley et al., 2016; Lawton et al., 2016; Bell et al., 2018; Pearce et al., 2018). However, even

outwith such scenarios, and trials, clinical pathways may create an unhelpful momentum. Noting how care processes are often structured to achieve time-to-treatment targets, Joseph-Williams et al. (2017) suggested they may encourage accelerated decision-making, including in situations where the impact of deliberative delay on clinical outcomes is likely to be very modest.

To wrap up, my finding that a range of features of the service context may shape patients' opportunities for, and experiences of, decision-making in powerful ways, complements an emerging body of work documenting the influence of policies, resources, demand management practices and clinical pathways. Calls for more attention to be given to these sorts of factors are growing (Scholl et al., 2018; Waddell et al., 2021). Critically, such important influences are unlikely to be accessible from talking to patients alone: my own research and that of others (McDonald et al., 2012; Vernazza et al., 2015) has demonstrated how the perspectives of direct care professionals can provide important, complementary insights. In addition, my work has highlighted the potential value of broadening the scope of enquiries (e.g., by interviewing other, *non*-direct care, professionals) to extend understanding of more distant structural influences.

Strengths and limitations of this work

In seeking to advance understanding it is important to acknowledge both the strengths and limitations of the work underpinning one's knowledge claims. Where, as here, such work has involved review and synthesis, shortcomings may derive from (either or both) the original research and the process of synthesis. In the paragraphs below I reflect on each of these in turn.

Considering firstly the original research grounding this submission, all my papers included detailed and (reflecting the changing expectations of editors and reviewers) progressively more structured Methods sections. The papers themselves highlighted a range of issues (with **Papers 4-7** including discrete 'Strengths and limitations' sub-sections). Identified strengths included:

- purposive approaches to sampling, to try and capture a diverse range of perspectives;

- use of semi-structured, sometimes serial, interviews, to explore topics in depth, and allow the emergence of issues unforeseen at the outset of studies;
- iterative approaches to data collection and analysis (with early findings informing subsequent data collection activities – both sampling and lines of enquiry);
- collection of data until 'saturation' was achieved; and
- 'member checking' (seeking feedback on findings from participants or their peers).

An additional strength is that though I led on and conducted the bulk of the work of analysis in each study, at least one colleague was always involved, providing challenge and confirmation. Such an arrangement is recommended in reporting guidelines (e.g., Tong et al., 2007) and increasingly a condition of publication.

Meanwhile, identified limitations included:

- achieved sample size and/or character (study 2, study 3), potential differences between those (professionals) agreeing and declining to take part in the study (study 3);
- co-production of (some) accounts, with (some) TYA and their parents being interviewed together (studies 1 and 3);
- one-off interviews, limiting ability to capture change, over time (study 3);
- reliance on retrospective accounts and potential for recall bias (study 3);
- study duration, more specifically the brevity of its follow-up period (study 2).

As all three studies sought to generate knowledge with the potential to change healthcare practice and/or policy, I prioritised dissemination to relevant (healthcare) professionals. My choice of journals reflected this, with all seven papers being published in clinical or hybrid journals. A defining characteristic of such publications is that they permit only relatively economical reporting of findings (**Papers 1** and **2**, for example, being limited to 3,500 words). An inevitable and unfortunate consequence was that scope for including illustrative quotes and contextual detail was limited – with obvious implications for the richness of both the original works and my synthesis.

Moving on to that process of review/synthesis, Noblit & Hare (1988) – arguably the pioneers of qualitative synthesis – advanced three criteria for evaluating work of this

sort. These were that synthesis should: clarify and resolve 'inconsistencies and tensions' in contributing work; bring about 'a progressive problem shift'; and be 'consistent, parsimonious and elegant' in character (Lee et al., 2015: 343). Numerous other criteria have been proposed as interest in qualitative synthesis has grown. Through extensive debate, some (though in no way complete) consensus has been reached about the desirable hallmarks of such work. Following trends in primary qualitative research, reporting guidelines/checklists have been produced for qualitative synthesis in general (the ENTREQ statement –Tong et al., 2012) and for more specific approaches to synthesis such as meta-ethnography (the eMERGe reporting guidance – France et al., 2019). Though the primary goal of such guidelines is to facilitate more complete and transparent reporting, as with comparable guidelines for reporting primary qualitative research (e.g., Tong et al., 2007) some assumptions about how the work (of synthesis) should be conducted are inherent. Indeed both Tong et al. (2012) and France et al. (2019) suggest that their guidelines may assist not only reporting, but also the design and/or conduct of syntheses.

Those guidelines place considerable emphasis on what might be termed boundary work: (a) articulation of a search strategy (either systematic or geared towards conceptual saturation); (b) the application of clearly defined inclusion/exclusion criteria; and (c) the conduct of some sort of quality appraisal activity, using a tool such as COREQ (Tong, 2007) or the Critical Appraisal Skills Programme's Qualitative Studies Checklist (CASP, 2002). With regard to my own synthesis these expectations appear of limited relevance: as the papers for inclusion in this critical review were determined on the basis of their authorship, no search was undertaken; and, appraisal was focussed on relevance and comparability, i.e., the feasibility of undertaking a synthesis, rather than quality *per se*. In defence of this approach, I highlight the more sceptical perspective on boundary work in synthesis which Sandelowski et al. (2007) expressed. They noted how the work of search, appraisal, and exclusion might be undertaken for different reasons, including to make a large body of literature more manageable (in terms of its scale and the ease of comparability) and/or to give work credibility (by adopting practices associated with systematic reviews). Whilst recognising both these aims as legitimate, Sandelowski et al. (2007:242) cautioned that over-diligent boundary work risked being 'so

exclusionary as to eliminate most of what constitutes the larger arena in which that phenomenon (of interest) is situated'. From such a perspective, the relatively inclusive stance I took may have acted to preserve and illuminate that larger arena (i.e., context). Thomas & Harden (2008) offered a complementary perspective, surmising that excessive homogeneity might discourage the abstraction and innovation essential to (interpretive) synthesis. Hence, they suggested, in undertaking synthesis some variability (e.g., in study settings) might usefully be sought.

Notwithstanding these arguments in favour of (some level of) heterogeneity, the work of synthesis *required* me to find areas of common ground in the publications in my portfolio. Turning again to Sandelowski et al. (2007:242), these authors observed that such 'comparability work' – or 'finding ways to work with or around study differences' is at the heart of the synthesis enterprise. They argued that as qualitative studies rarely deal with quite the same subject, much less in the same manner, managing diversity is a recurrent challenge. They argued, however, that such work has tended to be under-acknowledged, with (too) many decisions about which differences matter remaining firmly 'backstage'. So, to articulate the thinking and rationale underpinning my own choices, my three studies clearly varied in their aims, and decision-making was not in all instances their exclusive or even their primary focus. However, to varying degrees all progressed (my) understanding of and thinking about health-related decision-making. Having decided to include the seven publications, I did not exclude any content *a priori* from the synthesis, though I have only reported material here that, as a result of undertaking the work of synthesis, I deemed salient. Some of the seven publications contributed more than others to the final synthesis (i.e., the product), but all can be seen (Table 4, Appendix 2) to have furnished some piece of the jigsaw that constituted my final proposition, or line-of-argument.

My publications were not lengthy and detailed monographs: instead, as previously noted, several were prepared for journals with editorial policies requiring highly economical reporting. The necessary brevity of much contemporary reporting has been previously identified as presenting a challenge to synthesisers (Lee et al., 2015). Working with one's own material may be a benefit here, with familiarity with the datasets underpinning the published work facilitating insight. However, though

Noblit & Hare (1988) themselves sought to synthesise their own research, such an exercise might conceivably also have pitfalls, including the potential for blindness, due to over-familiarity with the work. Bondas & Hall (2007) noted how, in synthesis as with any form of research, there is the risk of finding what you expect or want to see; it is conceivable that this risk might be heightened when synthesising one's own work. I sought to guard against this, and check the urge to 'cut corners' in the process of synthesis, by imposing the discipline(s) of primary analysis (for example, line-by-line coding, constant comparison, mapping relationships between codes, themes, and proposition).

Other reflections on these undertakings

This leads me to consider, briefly, how, in other ways, my own position and the particular context of this work may have influenced either the process or product. As Hammersley (2004:9) noted, one's social location, cultural background, and prior beliefs (etc.) may assist or impede any given inquiry. Researchers, he advised, do not need to 'divest' themselves of all 'baggage', but should take care that this does not divert them from their course. Similarly, Maxwell (2012:97) asserted that researchers should 'take account of the actual beliefs, values, and dispositions that they bring' to a project, but these things should be considered 'as valuable resources, as well as possible sources of distortion'. This accounting seems eminently sensible, though somewhat easier in principle than in practice.

Personal characteristics such as my age, gender, class, education, language/speech, ethnicity, health, and domestic/familial status (e.g., childlessness) may, indubitably, have exerted (more or less subtle) influences on both how I approached (primary) data collection and/or how I subsequently analysed and interpreted my data. I recollect how with some research participants I had an awareness of shared characteristics, and on some level identified with them, whilst with others I experienced a profound sense of difference. It is hard to be sure which scenario is more or less problematic – as Murphy (1990:135) noted, even (others might suggest 'particularly') where researcher and researched share a defining characteristic (in his case/study disability), 'one must be careful not to assume that his own experience… is the same as his informant's.' Moreover, the potential effects of such personal characteristics may be amplified or muted by the conditions under

which research is undertaken (for example, how modestly work is resourced, and the degree of pragmatism this necessitates).

Pragmatic pressures and constraints are common to contemporary research, yet seldom acknowledged (being largely glossed over in published papers/reports). Tasked with meeting agreed aims and objectives with finite and rapidly diminishing resources (e.g., time) pragmatism is an absolute necessity in contract research, especially if one intends to publish. It was also an important consideration in preparing this critical review, which I undertook alongside my paid work. I should also declare a natural inclination to pragmatism, in the sense of being oriented more towards practical realities than theoretical ideals. This is evident in my general commitment to doing applied research, with the potential to lead to positive changes in practice and policy. Moreover, it informed this critical review via several decisions: not to adopt a distinctive disciplinary badge; to take a clearly operationalised (less cerebral) approach to qualitative synthesis; and to draw, in my discussion, on a bricolage of literatures from different disciplines.

Finally, the wider context of this work warrants some comment. In my Preface I noted that I undertook this review whilst the Covid-19 pandemic was underway, and the majority of research staff, including myself, were working from home. This situation meant that interactions with close colleagues were more limited that they might have been: various authors have noted the benefits of working in a team, in particular how dialogue with others in the research community can help to keep a researcher on course (Hammersley, 2004) and encourage the creative yet critical thinking necessary to achieve conceptual development (Glaser, 1965; Lee et al., 2015). I have of course had exchanges with my advisors and benefited from their intellectual input (in the case of Prof. Julia Lawton, input both to this review and four of the included publications). However, opportunities to test out ideas informally with other colleagues were restricted as a result of the pandemic and its impact on working lives. The work of synthesis would, I am sure, have benefited from the more extensive dialogic and collaborative work which underpinned the development of the papers themselves.

Implications for research and practice

The key message arising from this work is that what goes on alongside and outside consultations matters. This is not to negate the detailed scrutiny of planned clinical encounters and communicative practices undertaken by conventional SDM scholars, but to emphasise that it is also important to step beyond the consultation and consider the diverse contextual factors which may influence decision-making therein. With regard to future research, this shift in focus invites questions regarding those aspects of context of particular interest, and the tools and/or methods best suited to researching these. These concerns are of course related: as Bate (2014:20) noted, 'how we think about context will determine how we go about researching it'. In addition, it prompts reflection on whether and how current and future knowledge might have practical application, i.e., inform policy and practice.

Research considerations: what and how

I noted in my Introduction how, ahead of my own research, calls were growing for researchers to attend more closely to how family, partners and friends were involved in decision-making about treatment and care. This area of work has since received welcome attention, with both patients' and trusted others' perspectives being explored. However, methodological and practical challenges remain, which require attention if they are not to hold back understanding. As I reported in **Paper 1** (p1294) the influence of others 'may be significant without being obvious', and be exercised (substantially) outside the clinic. Notwithstanding researcher sensitivity, reflected in research questions, topic guides, and sampling ambitions, there are some barriers to capturing this influence via conventional interview studies. Firstly, such studies tend to require patients to recognise trusted others as in some way influential, be willing to talk frankly, and broker introductions so that researchers might involve those people in research. Secondly, there are substantial cost implications associated with gathering data via parallel interviews with trusted others: the realities of study budgets may not allow researchers to interview diverse samples of both patients *and* trusted others.

Other research methodologies and methods warrant consideration. Ethnography, for example, has been suggested as facilitating in-depth study of health-related phenomena within the context in which they transpire (Savage, 2000). The work of

Quaye et al. (2019) and Michinobu et al. (2021) on paediatric in-patients' participation in decision-making has shown how detailed and long-term observational work can illuminate the dynamic nature and influence of social context. However, ethnographic work is again costly (Savage, 2000) and undertaking this form of research with out-patients presents some obvious logistical challenges. As an alternative approach, work by Sanders et al. (2011) showed how analysis of posts to on-line discussion boards could provide interesting insights into off-line relationships and the management of illness within the context of daily family life, and highlighted how such posts differed in their immediacy and detail from retrospective interview accounts.

Without question, there is room for, and value in, further empirical work on how the physical and otherwise embodied experience of illness shapes decisions about treatment and care. However, the methodological challenges such work presents are again significant. As before, sensitisation to the issue may facilitate development of different and arguably better research questions and interview/topic guides. Nevertheless reliance on retrospective interviews, to collect data, may limit our capacity to understand such experiences (most particularly in respect of acute/severe illness – discussed further, below). Longitudinal study designs, whereby individuals' evolving experiences and decision-making processes are explored and captured via serial interviews (and/or observation) may extend understanding. But such designs are again costly; this has implications for the number and diversity of patients that can be involved. Alternatively, participatory and visual methods such as photovoice (Wang & Burris, 1997) might offer different insights into the experiences of certain populations (younger, more confident users of technology) and conditions (chronic illness and disability). Photovoice involves (research) participants taking, selecting, sharing, and reflecting on photographic images of relevance to a particular research topic/concern. This can highlight unexpected conditions, practices, and interactions, prompting new avenues of enquiry. The arrival and widespread possession of smartphones has expanded the accessibility of this method. However, new challenges are emerging: issues of privacy and information security are a growing concern for researchers and the ethics committees providing oversight for their work.

Capturing patients' experiences in more acute/critical phases of illness is likely to present ongoing challenges. Reflecting on his own experiences as a ventilated though largely conscious patient, Rier (2000) offered some suggestions, such as making use of records created for other purposes (e.g., the notebooks he used for communicating) instead of, in conjunction with, or as stimuli for retrospective interviews. Rier (2000) termed this practice (using patients' diaries or journals) 'inadvertent ethnography'. Such an approach has potential to provide quite unique access to embodied experiences; however, identifying patients who have and are willing to share such documents is likely to be a laborious process, and research findings (though rich) may ultimately rely on samples that are small even by the conventions of qualitative research.

Finally, more assiduous and scrupulous critical attention to the 'off-stage' processes and practices which frame and mould the content of clinical encounters would no doubt also be useful. In other fields/disciplines, such as implementation science and organisational change, interest in (service) context appears to be growing. I noted previously how, in study 3, the perspectives of professionals involved indirectly as well as directly in care provided important, complementary insights. These sorts of perspectives remain incompletely canvassed, and warrant future research. The potential of interview studies to progress understandings in this area remains, I believe, both substantial and underexploited.

Practice considerations

Perhaps the key question, with regard to practice, is how to acknowledge and take account of the various influences I have documented within consultations. The 'Discussion' sections of the individual papers offer a number of reflections and proposals specific to the populations, scenarios, and services on which they are focussed. Here, I draw out some broad, higher-level considerations, relating to each of the three contexts which this document has explored:

- Personal context*:* There are times when the direct – embodied – experience of illness (and indeed treatment) makes it almost impossible to absorb, process and act upon information provided for the purposes of decision-making in any meaningful way, and others when patients struggle to look beyond resolving very short-term concerns. Where patients express preferences for a more directive

approach this should be respected, but not assumed to be a permanent orientation. Receptivity to – indeed desire for – information may change, in line with (amongst other things) changes in embodied experience. The goals, risks and benefits of treatment should therefore be revisited periodically, with opportunities provided to discuss (new) priorities and concerns as and when these emerge.

- Social context*:* The involvement of 'others' is normal. People around patients may play important though not necessarily obvious roles in decision-making, facilitating deliberative activity, extending or constraining patients' (sense of) having options, and enabling patients to follow-through on the choices made. However, who these people are, and the role they play, may vary both between individuals and over time. Some patients, at some time points, will lack the support that others take for granted; in certain instances the involvement of others may not be entirely benign. Exploration of individuals' current circumstances and concerns is therefore key, including identifying who else they might want involved in decision-making and how. The provision of information, and organisation of care, can then take the role and influence of those people into account. This recommendation is consistent with very recently updated UK guidelines on SDM (National Institute for Health and Care Excellence, 2021) which advocate: checks on whether patients want to involve others (and who); and the offering of additional support (e.g., volunteer advocates) to patients who do not have, or want, support from family or friends.
- Service context: The organisation of care and research, and resource frameworks underpinning this, also has implications for the choices patients (may) make. Both the actual and perceived resource implications – the 'economics' of care and research – influence health professionals' decisions and behaviours, and through this effectively determine the decisions/options available to patients, for instance around participation in clinical trials. Systems of rewards/incentives and 'penalties' inform and shape professionals' practices – it is important that policy-makers understand their effects and, inasmuch as financial realities allow, are ready to make changes where such systems constrain choice unintentionally and unnecessarily.

REFLECTIONS ON METHODOLOGICAL DEVELOPMENT AND LEARNING

In this final chapter I consider: my experiences of becoming and being a contract or 'early career' researcher; what I have learnt through from my experience of undertaking different research studies; how my thinking – and approach to both research and writing – has evolved over this time; and what I have gained from synthesising earlier work and preparing this critical review.

Becoming and being a contract researcher

Whilst life as a contract researcher has its downsides – most obviously occupational insecurity – it also has many positives, amongst these variety in, and scope for learning from, changing topics, tasks and teams. This point is well illustrated by the three studies from which my synthesised publications arose, which related to different clinical areas and populations, involved different academics and clinicians at different institutions, and though all in the qualitative tradition, had somewhat different research designs.

Learning from different research studies

The three studies whose findings I report in this review all provided valuable opportunities for learning. I have distilled out the following important, pragmatic lessons:

- ***Realism in recruitment:*** The success of qualitative projects relies on one's ability to recruit, yet difficulties here are common. Whilst some challenges may be inevitable, others might be foreseen and, to an extent, mitigated. For example, I have observed a tendency amongst enthusiastic (clinician) investigators to overestimate their own (or colleagues') capacity (and willingness) to recruit. Protocols using the 'academic-sounding' passive voice now ring alarm bells, as this conveniently obscures the detail of who will actually do what, when clarity on this point is essential. Recruitment processes need to be very clearly articulated (who exactly will identify and approach potential participants, how information will be shared if different people are involved) along with assumptions about eligible patient populations (numbers, distribution) and access to tools for their identification (e.g., specialist databases) plus informed assessment made of the challenges and opportunities for securing relevant professionals' support. Where

these things have not been adequately thought through, changes to the research plan may be required, with these generally involving further applications for ethics/governance approval (see below) and additional, un-resourced, on-the-ground work by the project researcher.

- ***Recognising the costs of change:*** One of the great strengths of qualitative work is its emergent and flexible approach to research design and delivery. Some changes – for example, to the detail of an interview guide/schedule – can be accommodated within the general plan/protocol. However, others require review and approval by the research sponsor, research ethics committee (REC), and NHS Research and Development (R&D) committee. In all three studies it proved necessary to seek and secure approvals for one or more 'minor amendments' to the study plan. Considerable amounts of my time, as well as that of other professionals, were spent securing approval for relatively modest changes, e.g., to recruitment processes and mediums for data collection. Hence, whilst I would strongly favour maximum clarity within research teams – and critical thinking about – envisaged processes and procedures, I would also suggest that (initial) ethics and governance applications provide no more potentially constraining detail than is absolutely necessary. Where changes to project plans are necessary, the associated costs – including the likely impact on time available for advanced analysis and writing for publication – need to be properly recognised.
- ***Prioritising publication:*** Perhaps the most critical lesson I have learnt is the importance of keeping study aims and outputs in clear sight, beginning analysis early, recognising when sufficient data have been collected (accepting that there is a trade-off between collecting more data and investing time in its analysis) and allowing adequate time – within the project's lifespan – for writing up and otherwise disseminating study findings.

Thinking about research (and writing)

In introducing this review, I explained how my substantive thinking had evolved, with its development being characterised by an increasing interest in, and attention to, what might be broadly labelled 'context'. I now note how alongside – and in conjunction with this – my thoughts on matters of epistemology and methodology (what we can know and how we might know it) also matured and crystallised. As indicated in the submitted papers, influences upon my thinking about, and practices

of, research and/or writing have at different time points included: (classical and more constructivist) grounded theory (Glaser, 1965; Glaser & Strauss, 1967; Strauss & Corbin, 1990; Charmaz, 2014); qualitative description (Sandelowski 2000 & 2010); and more recently critical realism (Maxwell, 2012; Alderson, 2021). One of the distinctive features of academic writing – by which I mean writing for peer-reviewed publications, rather than producing a research report – is the expectation that authors articulate their position and practices, using labels which have meaning and currency in their field. Characterising one's orientation and work in this way may be helpful and add transparency – though a decade of reading academic papers suggests this is not guaranteed. Different epistemological and methodological labels or 'badges' seem to be applied to remarkably similar undertakings and, not infrequently, suggest features/characteristics which a given piece of work does not – at least as reported – seem to manifest.

Returning to my own work, and the worldview underpinning it, the various influences cited in the included papers reflect less a marked change in outlook and practice, and more a change in my capacity to articulate what that underpinning worldview is and how it informs my work. More specifically, they illustrate a shift from an implicit – to explicit – alignment with critical realism, broadly in the form described by Maxwell (2012). This – as noted in **Paper 6** (p2) – is 'a philosophical orientation combining a realist ontology and constructivist epistemology', or the position that there is an external reality, which exists independently to our perceptions, but (the 'critical' bit) our understanding of it will always be partial, situated and socially-fashioned. Critical realists view these concerns (things and our perceptions of them) as distinct, though equally real, and 'mutually influencing' (Maxwell, 2012: viii). Of particular importance to this piece of work, is that critical realism revives and re-legitimates interest in ontological concerns, i.e., the nature of phenomena as well as the meaning people attach to them, and recognises context as important, influencing beliefs and behaviours and having a central role in explanation. Critical realism is a position broadly supported and/or endorsed by several influential qualitative researchers and social scientists (Miles & Huberman, 1984; Hammersley, 1992; Hammersley, 2004) and – notwithstanding vocal advocacy for a more constructivist (and relativist) ontology by some colleagues – a 'common sense realist ontology' characterises much contemporary qualitative research (Maxwell, 2012:6). Fundamentally critical

realism is pragmatic and Maxwell notes how ‘it does not discard *a priori* those approaches that have shown some ability to increase our understanding of the world’ (Maxwell, 2012: 10). Qualitative synthesis is, therefore, an endeavour that fits comfortably with a critical realist outlook.

Experience of and gains from preparing this critical review

As Maxwell (2012) has noted, we both influence and are influenced by what we study. Both the primary research and my work on its synthesis have had an effect on me as a researcher (and as an individual). Historically, the organisation and resourcing of contract research has encouraged me to ‘draw a line’ under old projects as soon as possible, consigning them to the past in order to focus on a new brief and meet new milestones. To an extent this is necessary, to free up time and cognitive capacity to grapple with new – and often quite unfamiliar – topics (with these, in applied health services research, often relating to a previously unknown and complex area of medicine). However, the process of undertaking this review has shown me how returning to, and reflecting upon, a body of past work can lead to new understandings and insights.

Whilst each project, and paper, sought (in some cases amongst other things) to inform understanding of some aspect of patients’ decision-making about treatment and care (or in the case of study 2, self-care), bringing the work together has enabled me to develop, and offer, a more nuanced and powerful line-of-argument. The need to situate my findings incentivised me to read (much) more deeply and widely, including exploring the literature of disciplines/fields such as psychology, emotion science, and bioethics, with which I previously had little familiarity. The review process further prompted me to draw out, and reflect upon, the strengths, weaknesses, and opportunities for learning provided by each study; highlighted continuing gaps in the evidence base; and, sensitised me to important questions and methodological considerations. I expect these things to inform my work around health-related decision-making, and other aspects of patients’ experiences, going forward.

CONCLUSION

In closing I seek, as per Wolcott (1990), to summarise succinctly what I have attempted and achieved in this review. Fundamentally, with the aim of advancing understandings of health-related decision-making, I set out to situate, present, draw together and critically consider relevant findings from my own 'back catalogue' of publications. To bring together my work as a coherent whole, I employed techniques of qualitative synthesis – a practice which, as Thorne et al. (2004:1346) contended, 'can be understood as a form of discourse that contributes to a fuller understanding of the phenomenon of interest'.

My synthesis enabled me to develop and evidence the proposition that ***health-related decision-making happens in and is shaped by context***. Specifically, I highlighted the importance of three particular kinds of context – the personal, social and (health) service contexts of decision-making – and documented three associated analytic themes. In doing so, I have built upon and extended the work of sociologists with interests in SDM, including that of my former colleague (and PI) Rapley (2008). Importantly, I have raised questions about the adequacy of SDM scholars' longstanding focus on what goes on within the boundaries of clinical encounters.

Whilst what goes on in consultations clearly matters, my review indicates that to fully understand decision-making, we also need to consider the complex contexts in which it takes place. As yet this need appears under-recognised (both under-researched and under-theorised) notwithstanding the recent explosion (Lu, 2019) in SDM-related publications. Indeed, Thomas et al. (2021:2) very recently lamented how prevailing models of SDM remained essentially mechanistic, privileging 'reasoned deliberation' whilst neglecting relational elements including affective, cognitive, and sensory experiences, as well as the broader cultural and 'environmental' contexts in which clinician-patient encounters took place. My work, I posit, offers a timely and powerful corrective.

On a subsidiary note, it became clear in situating my work that other, disparate literatures can offer useful and (to date) under-exploited insights. Excepting the work of Rapley (2008), relatively limited use has been made (by SDM scholars) of relevant writings originating from within medical sociology. SDM scholars also appear to have

paid surprisingly little attention to salient research emanating from psychology and emotion science. It is yet to be seen if emerging literature from the fields of service design and implementation science will be taken into account: these bodies of work look to have considerable potential to improve understanding of organisational and/or service level influences on SDM. Ultimately, the case made by Ferrer & Mendes (2018) for greater ‘crosstalk’ between disciplines with cognate interests is a sound one, for which my review provides strong support.

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APPENDIX 1 (ADDITIONAL THEMATIC MAPS)

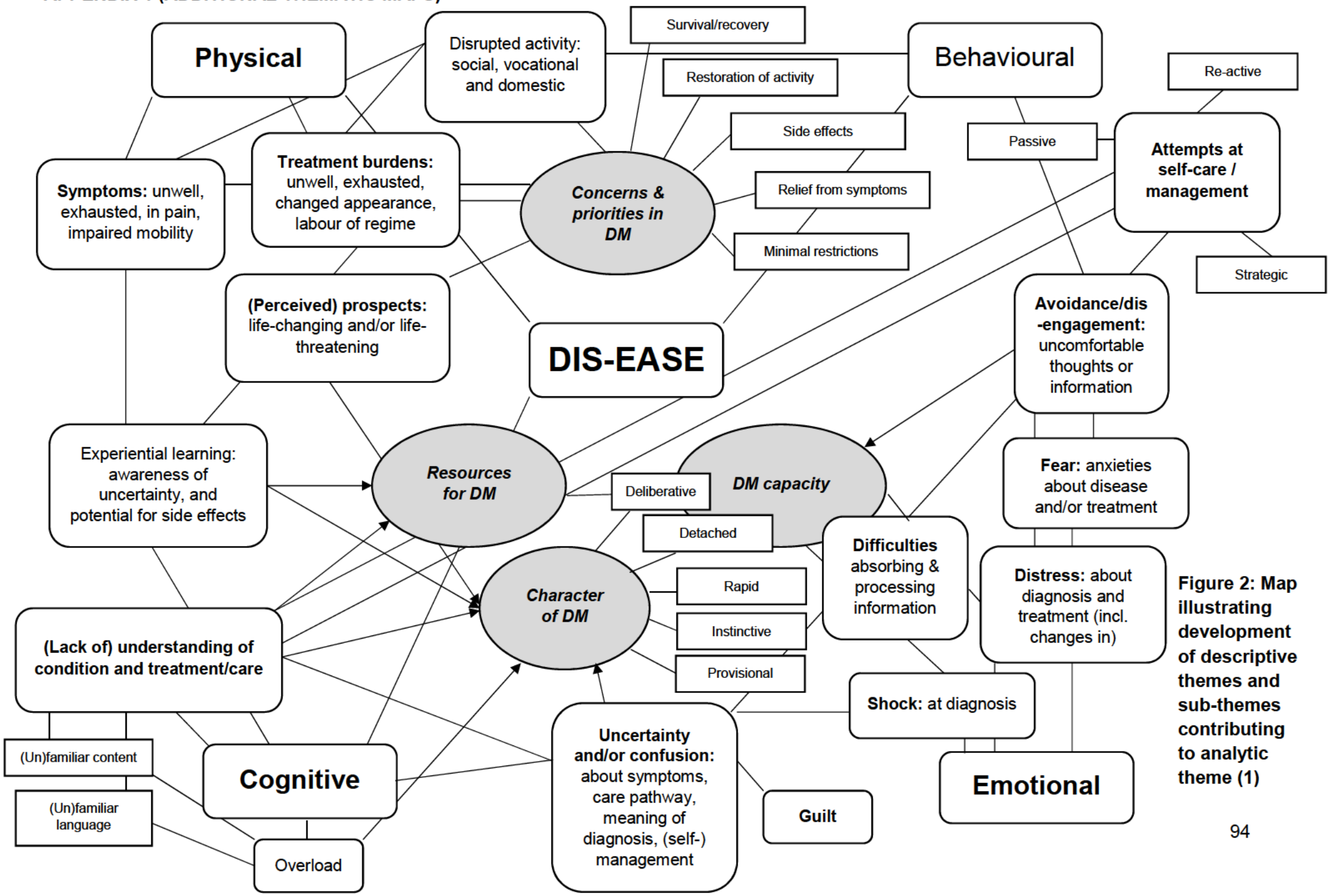

Figure 2: Map illustrating development of descriptive themes and sub-themes contributing to analytic theme (1)

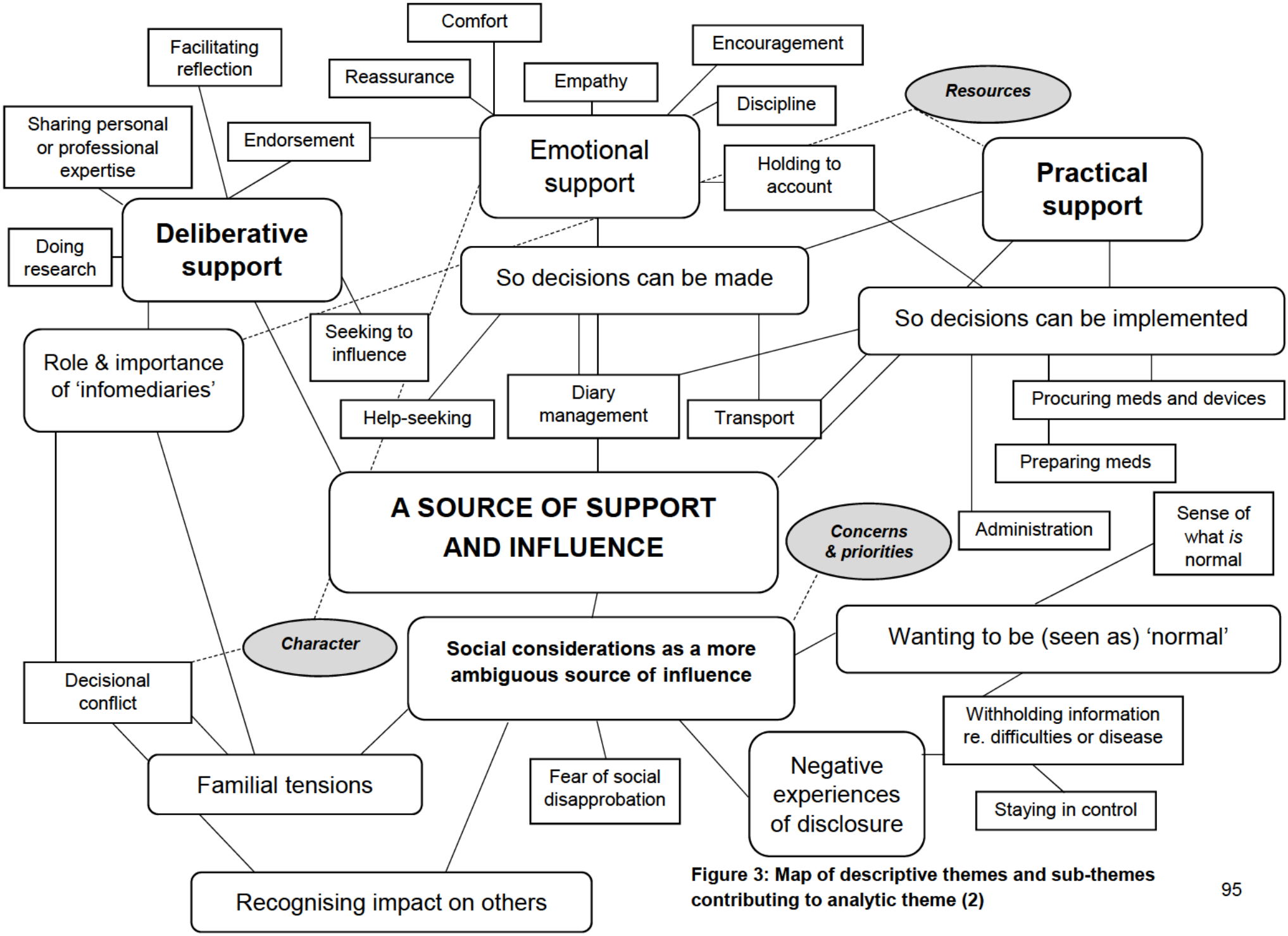

Figure 3: Map of descriptive themes and sub-themes contributing to analytic theme (2)

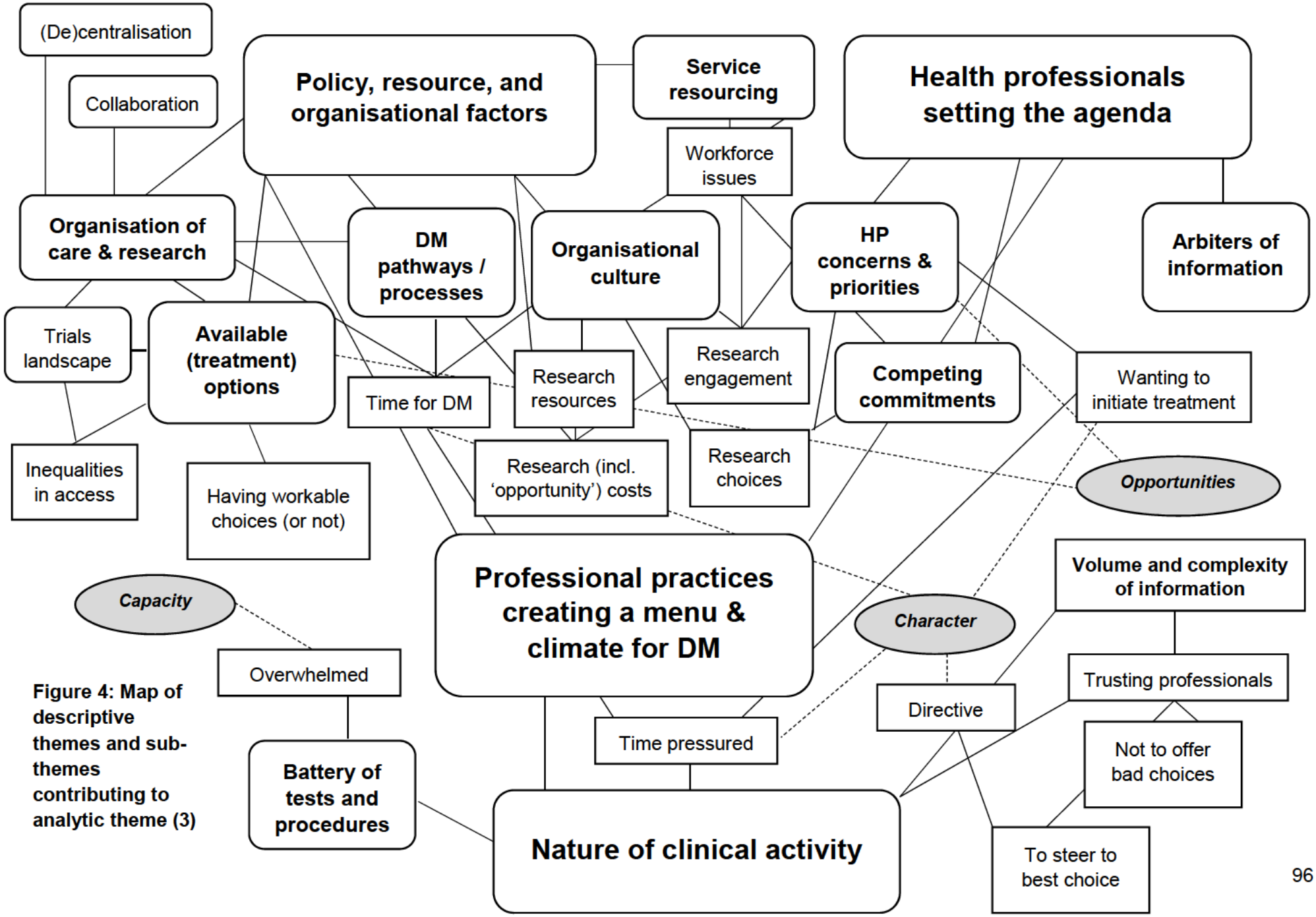

Figure 4: Map of descriptive themes and sub-themes contributing to analytic theme (3)

APPENDIX 2

			Study 1		Study 2	Study 3			
			Paper 1	Paper 2	Paper 3	Paper 4	Paper 5	Paper 6	Paper 7
Health-related decision-making happens in and is shaped by context	Personal context: experiencing a state of dis-ease	Physical dis-ease		X	X		X		
		Emotional dis-ease		X	X		X		
		Cognitive dis-ease			X		X	X	
	Social context: a source of support and influence	Deliberative support	X	X	X		X	X	
		Practical support	X				(X)	(X)	X
		Social considerations	X	X			X	(X)	
	Service context: creating a menu and climate for D-M	Policy, resource & org. factors		(X)		X			X
		HP setting consultation agenda		X	X				
		Nature of clinical activity					X	X	
Note: ( ) indicate a more minor contribution									

Table 4: How each study and paper contributed to the synthesis

APPENDIX 3 (SUBMITTED PUBLICATIONS, PAPERS 1-7)

RHEUMATOLOGY

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Original article

Young people's decisions about biologic therapies: who influences them and how?

Ruth I. Hart[1], Helen E. Foster[2], Janet E. McDonagh[3], Ben Thompson[4], Lesley Kay[4], Andrea Myers[5] and Tim Rapley[1]

Abstract

Objectives. Young people with inflammatory arthritis can have severe disease warranting biologic therapy. They face complex treatment decisions, with profound consequences. This study aimed to explore the influence of individuals outside the care team (trusted others) on the treatment decisions made by young people, in particular their decisions about biologic therapies.

Methods. Young people (16–25 years of age) with inflammatory arthritis and experience of treatment decision making were recruited from three NHS Hospital Trusts. Twenty-five were interviewed, plus 11 trusted others identified by young people as being involved in their decision making, as well as 6 health professionals. The data were analysed using coding, memoing and mapping techniques and the findings were tested through a series of focus groups.

Results. Young people initially emphasized their decisional autonomy, typically describing people other than health professionals as limited in influence. However, discussions revealed the involvement—in deliberation and enactment—of a range of other people. This cast of trusted others was small and largely consistent; mothers played a particularly prominent role, providing cognitive, practical and emotional support. Members of the wider cast of trusted others were involved in more limited but still significant ways.

Conclusion. Young people claim autonomy but other people enable this. The network of relationships in which they are embedded is distinctive and evolving. Mothers play a supporting role well into early adulthood; in contrast, partners are involved in far more limited ways. As such, the applicability of adult models of decision making is unclear. This must be taken into account if the support provided by professionals is to be optimally tailored to young people's needs.

Key words: young people, inflammatory arthritis, decision making, biologic therapies, trusted others, interdependence, qualitative research.

CLINICAL SCIENCE

Rheumatology key messages

- Young people claim autonomy in treatment decision making, but other people, especially mothers, enable this.
- Other people's influence on young people's treatment decisions may be significant without being obvious.
- In promoting independent decision making, care teams should take relationships with trusted others into account.

Introduction

Recent years have seen important changes in the clinical management of inflammatory arthritis, in particular the widespread use of biologic therapies in both paediatric and adult services. Young people with aggressive disease are more commonly being offered such treatments, and at an earlier stage in the disease course [1]. The evidence is clear that short-term benefits can be considerable, and include reductions in joint pain and damage, plus

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improved mobility. However, there are also short-term risks (e.g. increased vulnerability to infection), and the long-term consequences of these treatments (e.g. impact on fertility, risk of malignancy) remain uncertain [2]. This is of particular concern for those who begin taking them early in life. Young people offered biologics are therefore confronted with a decision that may have profound consequences, at a point when their disease is at its worst and their wider lives are characterized by change and uncertainty.

Health care professionals play an important role as providers of information and advice for patients generally [3] and for young people specifically [4, 5]. However, treatment decisions have also been shown to be influenced by interactions with people outside the health care team [6, 7]. In considering lay influence, the research literature focuses substantially on the significant other, with this term typically connoting a long-term partner or spouse [8, 9]. Yet in the UK the trends are clear: people are committing to a partner much later than in the past [10, 11]. If a growing proportion of young adults do not have a significant other, focussing exclusively on this relationship as a source of influence or support for decision making is problematic. We therefore looked more broadly at the who, how and why (or why not) of lay involvement in young people's treatment decisions. We refer to this broader group of people as trusted others.

Methods

We report here on one component of a wider study of young people's decision making regarding biologics. That study employed a range of qualitative methods: interviews (with young people, trusted others and health professionals), recording of patient/professional interactions and focus groups. The analysis reported here draws principally on data from interviews, but is informed by learning from other study strands and was validated in the concluding focus groups. The study conformed to National Institute for Health Research requirements and had Research Ethics Committee approval from the Proportionate Review Sub-committee, National Research Ethics Service Committee Yorkshire & Humber—Leeds East (ref. 12/YH/0122). All participants gave consent verbally and in writing.

Setting

Potential interviewees (and participants in other research strands) were identified and recruited via three NHS Hospital Trusts, two in the North East of England and one in the West Midlands. These trusts operated one or more of the following rheumatology services: adult clinics, young adult clinics run by adult and/or paediatric rheumatologists with interests in adolescence and adolescent clinics run by paediatric rheumatologists.

Sample

Our approach to sampling was purposive, seeking to encompass variation in demographic characteristics, diagnosis and treatment history (see Table 1) and to explore emerging conceptual issues. Requests were made to direct care colleagues to identify and approach young people with specific characteristics. Young people ($n = 25$) were between 16 and 25 years of age at the first interview and had a diagnosis of inflammatory arthritis (either JIA, AS, PsA or RA). At first contact they either had not yet been offered a biologic ($n = 5$), had recently been offered a biologic ($n = 5$) or already had some experience with one or more biologics ($n = 15$). Where young people's treatment status changed, i.e. as they started taking a first or subsequent biologic, attempts were made to re-interview them.

Trusted others ($n = 11$) were identified by participating young people and approached through them. Most agreed to participate and this subsample of interviewees included eight mothers, one father, one grandmother and one partner. Trusted others who declined to participate included a close friend and a partner. Health professional interviewees ($n = 6$) were identified by the core research team and chosen to include key roles within the multidisciplinary teams at the participating trusts and their service providers.

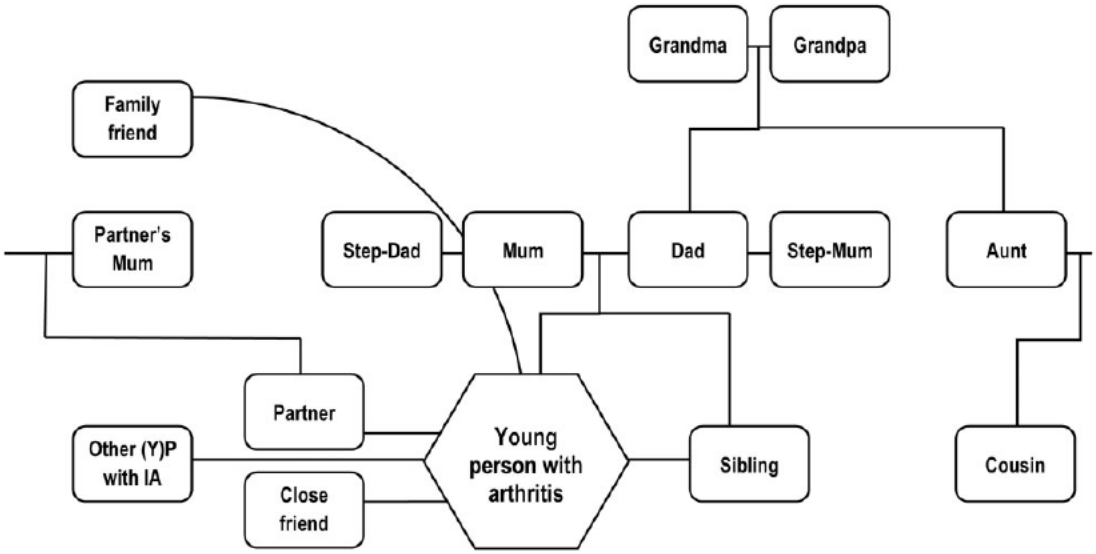
Table 1 Characteristics of young people interviewed

Characteristic	Value
Diagnosis, *n*	
JIA	15
AS	7
PsA	2
RA	1
Gender, *n*	
Female	15
Male	10
Age, mean (range), years	20 (16–25)
Disease duration, mean (range), years	9 (<1–>20)
Rheumatology service accessed, *n*	
Adult clinic	10
Young adult clinic	8
Adolescent clinic	7

Data collection

R.I.H. interviewed 25 young people, 5 on more than one occasion, plus 11 trusted others and 6 health professionals. None of the interviewees had encountered R.I.H. prior to the start of the project. In five cases, young people and trusted others were interviewed together; in all other instances interviewees were spoken to individually. Interviews were semi-structured, lasted 40–120 minutes and were predominantly conducted face to face at a location of the participant's choice. Interview schedules were initially informed by the team's experience and a review of the literature. These were adjusted to take account of individual circumstances (e.g. young people's treatment status) and refined following each round of analysis. All interviews were recorded, transcribed verbatim

FIG. 1 Trusted others featured in our data set



and anonymized. Field notes were written after each contact and provided an additional resource for analysis.

Data analysis

The study data comprises 52 transcripts (of which 44 relate to interviews). These were closely and systematically examined by R.I.H. using open and focused coding, mapping and memoing techniques to identify, classify, label and relate themes, phenomena and ideas [12, 13]. Data segments (selected transcripts or data pertaining to a particular theme) were similarly analysed by T.R. Analyses were compared, shared and developed further with other researchers in fortnightly data clinics, biannual team and steering group meetings and a concluding series of focus groups. These four focus groups comprised young people ($n=7$, $n=3$), trusted others ($n=4$) and health professionals ($n=8$). They were a vehicle for establishing face validity, providing a forum in which research participants and their peers could comment on the intelligibility, credibility and significance of the findings.

Results

My decision . . . but: stories of enabled autonomy

In the following sections we report on the involvement of others in young people's treatment decision making and look in detail at four important roles these trusted others play. Overall the message is one of qualified autonomy, encapsulated by the recurrent expression 'my decision . . . but'. This echoes research into young people's experiences of treatment decision making in other clinical areas [4, 14, 15]. However, while the findings of those studies suggest constraint, the stories emerging from our data were largely about enablement.

In general, young people in our study began by emphasizing their autonomy, typically describing people other than health professionals as limited in influence. However, in subsequent discussion they revealed the involvement of a number of other people in the making and making possible of treatment decisions. A mapping exercise (Fig. 1) showed this cast of individuals with influence to be relatively small and largely consistent. All relationships had foundations in the real world and the group was dominated by close family. Mothers played a particularly prominent role in the accounts, as detailed in the following subsection.

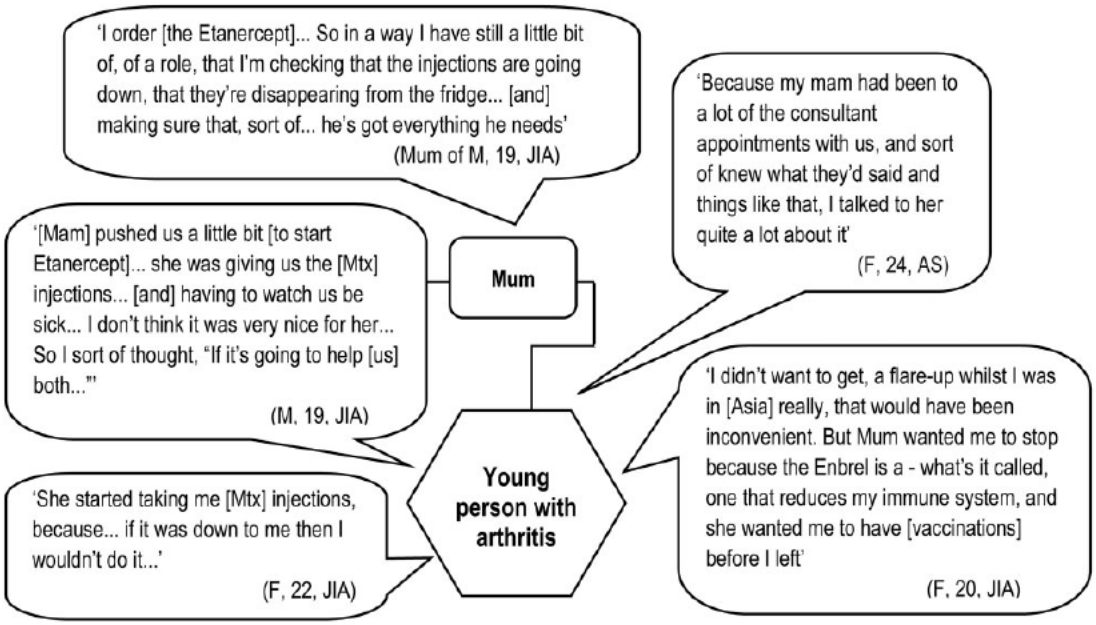
Trusted (m)others: I've always had me mam there

Mothers featured prominently in stories of making and enacting decisions in around three-quarters of cases. The majority of these young people (17/20) were living under the same roof as their mother at the time of the research. In two further instances, where the young person's mother was not able to play such a significant role, someone else (a father and a grandmother) had stepped in. The small minority of young people who did not acknowledge the role of a mother, or stand-in, had distinctive characteristics, typically having adult-onset conditions that had been diagnosed after leaving home and/or moving in with their partner. In essence, these were young people who were organizing their lives like adult patients.

Mothers are distinctive for the centrality of their role and for the variety of ways in which they are involved in decision making. It is common for them to be implicated in both deliberation and enactment, and their involvement spans practical, cognitive and emotional realms (see Fig. 2).

Examples of practical support include facilitating access to services and enabling (sometimes enforcing) the following through of decisions made in clinic. Young people and trusted others explained how mothers were

FIG. 2 Mothers' involvement in and influence on treatment decisions



involved in some or all of the following tasks: making appointments; taking the young person to the hospital; ordering, receiving and storing medications and preparing and in some instances administering injections. It was exceedingly rare for anyone else to be involved in the more intimate of these tasks (e.g. administering injections). Young people understood that treatment regimes impacted on their mothers as well as themselves; a change in treatment was therefore seen by some young people as having the potential to help them both.

Mothers were also portrayed as supporting and contributing to the cognitive aspects of decision making. This included prompting or asking questions, doing research, providing information and checking understanding. In many instances they accompanied the young person to consultations—as such they provided a backup or aide-mémoire and were uniquely positioned to discuss the pros and cons of the option(s) presented to the young person. They also acted as a conduit, relaying information to other members of the family on the young person's behalf.

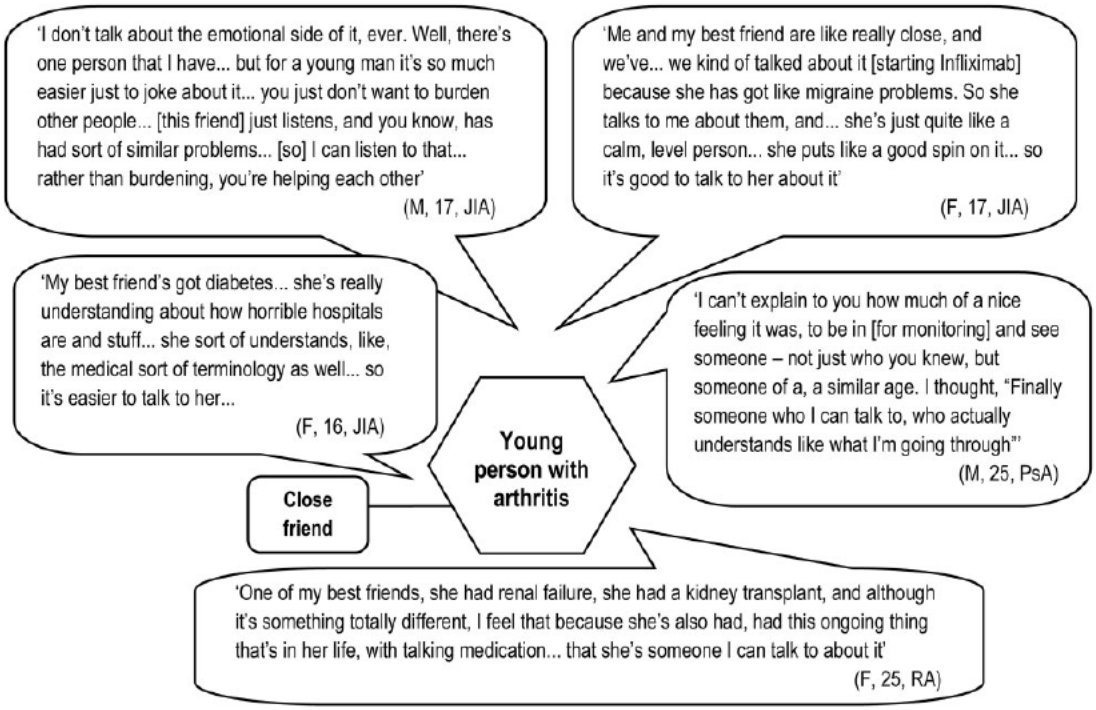
Mothers additionally offered emotional support to confront an important decision at a difficult time, essentially being there for the young person and providing reassurance, comfort and encouragement. However, young people's choices regarding where to turn for emotional support were complex. They sought at times to protect their parents (who often felt responsible for decisions made, delays in diagnosis and sometimes the condition itself), looking elsewhere to meet their emotional needs.

Wider family: in-house experts

Members of the wider (non-nuclear) family appear to have defined but decisive roles when, in addition to ties of blood or marriage, they can claim relevant professional expertise. For example, young people talked of step-parents, an aunt and a cousin who were nurses, or allied health professionals. Young people often viewed this in-house expertise as a valuable resource. These extended family members typically had limited practical involvement, but were providers of information and advice about both the young person's condition and potential treatment options. As such, they were in a position to substantially and directly influence treatment decisions.

A young person (male, 25 years old, diagnosis of AS) first interviewed in the early stages of the project provides a good example of this. This young man had recently transferred into the Trust, though he had had the condition since his late teens. He had met his consultant just once at the time of the first interview and was taking NSAIDs. Biologics had not been discussed. However, in the interview it emerged that he already knew about them and hoped to discuss these treatments with his consultant at his next appointment. Further questioning revealed that he had learnt about this group of drugs from his cousin, the young man explaining: 'My cousin, she's a nurse . . . she'd actually written down some names of drugs to suggest . . . So she's had a bit of an influence too'. A few months after that interview he saw his consultant again and subsequently began taking adalimumab. A second interview was arranged in order to explore the circumstances of the treatment offer and decision. It transpired that (like several other interviewees with the same diagnosis) he had been given a choice of biologic. The young man explained his decision as follows: adalimumab was 'the one I was recommended from my cousin . . . so I just plumped for that'.

FIG. 3 The importance of friends with similar problems



Empathic friends: someone I can talk to

Offering a quite different—but still highly valued—kind of expertise are friends (or family members) with personal experience of ill health. These people influence orientation to new treatment options and provide support to manage the emotions associated with starting new regimes with uncertain outcomes. About a third of the young people in our sample identified one or more friends of a similar age (occasionally their boy-/girlfriend) who had their own direct experience of ill health and with whom they felt able to share quite intimate details of their illness experiences and treatment dilemmas (see Fig. 3). These friends had a variety of conditions, including kidney failure, diabetes, arthritis and migraines. They demonstrated three important qualities that set them apart from interviewees' other peers. First, these friends had a more grounded understanding of their experience and capacity for empathy. Secondly they had knowledge of the organization of health services and medicine. Thirdly, their own difficulties provided opportunities for reciprocity. This sense of being able to help, rather than burdening each other, was clearly important to the young people in our study; this accords with the emphasis of other authors on reciprocity as the key component of friendship [16].

In contrast, young people appeared hesitant, and highly selective, regarding discussing either their treatment or condition with their wider/healthy peers. Some admitted this quite frankly. Others had their claims of being open about their condition challenged by a trusted other, subsequently qualifying their account. When probed, concern was expressed about how peers would respond. Some young people already had negative experiences of disclosure; others anticipated these. It was important to young people to be seen as normal, not as different in any significant way from their peers. However, they also talked of struggling to convey information about their condition and other people's struggles to understand. This inability to comprehend the nature of their condition made the idea of involving healthy peers in deliberations about treatment and care look quite pointless.

Supportive partners: someone to put the foot up your backside

Almost half of the young people interviewed mentioned a boy-/girlfriend, fiancé(e) or spouse, with six saying they were living together. However, partners rarely featured prominently in young people's accounts of treatment decision making. No examples were found of partners providing practical support (e.g. with receipt, storage, preparation or administration of medications); where this was needed, young people turned to their mothers. Cognitive involvement also appeared modest, with young people using their partner, at most, as a sounding board. Where partners' involvement was consistently reported was as providers of encouragement, motivation and discipline. A trusted other, whose son had lived away from home for some time, commented that people tend to get lazy with their health and the attention of someone who cares is of real value. Regarding his own son he said, 'His partner now is, you know, nagging him more than I did—and he takes it better!'

Although young people typically said their partner knew about their condition, what that meant in practice seemed to vary considerably. It was rare for them to take their partner to hospital—just one of our interviewees reported that her boyfriend routinely attended consultations. Another said they would welcome their partner along, and some suggested they might involve their partner further if and when they considered starting a family. Most, however, expressed a clear preference to see the consultant alone, allowing them to manage the information their partner received and maintain control over any decisions made.

A few young people said they had been encouraged by health professionals to share information with their partner. They had found this advice valuable, if hard to follow. Several admitted to playing down their difficulties, saying they did not want to be fussed over or to let their partner down. So, on balance, partners appear relatively poorly informed. Fluctuations in a young person's health, treatment regimen and responses might be observed, but are far from fully understood.

Discussion

Young people with inflammatory arthritis who are confronted with decisions about biologic therapies vary along multiple dimensions. In addition to demographic variation, significant differences are evident in their disease trajectories and treatment histories. Despite these differences, however, we see commonalities of experience that draw them together as a group and set them apart from the more typical (older) rheumatology patient.

It is clear that while young people claim decisional autonomy, and a small minority are justified in doing so, most exhibit a relational autonomy. Their autonomy is enabled by others who shape and support the making and enactment of decisions. This is true for adults too—the literature suggests that autonomy is enabled across the life course—but different people are involved, in different ways [6, 7, 17, 18], and critically, attitudes towards their involvement are different.

Our study found that mothers often remain involved in a wide range of ways well into early adulthood, in particular—but not exclusively—where their child is diagnosed while a child. Young people in whose stories mothers (or a stand-in from within the close family) do not play a prominent role were a small and distinctive minority in our study. All had adult-onset conditions, but in addition had been diagnosed after leaving home and/or starting to cohabit with a partner. These were young people who were organizing their wider lives in an adult way.

Partners replaced parents as the first port of call in a yet smaller minority of our cases (a situation noted in other recent studies [19]). The role they take on is typically much narrower than that of mothers, and careful management of information, or partial disclosure, appears the norm. In line with previous research on disclosure (of genetic risk) in dating [20], our data suggest that sharing information with partners is seen as risk-laden and difficult. As previously pointed out elsewhere [21], insufficiency of information can cause relationship tensions and lead to misguided support for patients. Help to think through whether, when and what information to share with their partners (and indeed healthy peers) might benefit some young people both emotionally and, ultimately, clinically.

While relationships with healthy people are important [22], friendships with other people with chronic illnesses were highly valued by the young people in our study. On occasion their experiential knowledge directly informed treatment decision making. These relationships also appear to have wider and potentially lasting benefits [23, 24]. Friendship choices are rarely the preserve of individuals alone [25], hence the position taken here by care teams (and others) is important. The case for recognizing and the potential for facilitating the development of friendships with other young people with inflammatory arthritis, or chronic illnesses more widely, is worth exploring further.

The involvement of others is normal, not dysfunctional, and for patients in other age groups is largely accepted, if not entirely approved. Young people need staunch allies, and for many of the young people in our study (as suggested elsewhere [26]) their mother continues to be the best candidate. However, unlike older patients, young people are encouraged, if not required, to demonstrate independence in various ways [19]. While recognizing the importance of work to empower young patients, we believe considerable care needs to be taken to promote independence without forcing supportive relationships underground. We acknowledge that paediatric teams are increasingly working towards interdependence, where young people take responsibility for themselves but parents continue to function as consultants [27].

Our data also offer a reminder that young people cannot rely equally on their parents for support and guidance. They may be disadvantaged by family structure [28] or by resources [29]. Simmons *et al.* [15] draw our attention to young people within the care system who may have autonomy forced upon them. These, and other young people whose families are struggling or fractured, may benefit from additional professional attention and support.

Critically, interactions outside the clinical consultation matter, but while the patterns highlighted here provide a prompt to question received wisdom and taken-for-granted practices, they do not tell us who influences treatment decisions, and how, in any particular case. Hence exploring home and peer relationships using screening tools such as the Home, Education/Employment, Activities, Drugs, Sexuality, Suicide assessment tool [30] should be routine practice for all young people.

Being clear who is involved, and how, is important for several different reasons. First, it is the only way to make sure everyone involved has appropriate information—something that Elwyn *et al.* [31] have argued is a fundamental component of effective shared decision making. Secondly, such clarity equips the health care team to foresee challenges and pre-empt potential

problems (e.g. on the young person moving away from home). Finally, it will help professionals identify the need for and opportunities to build independence at a pace appropriate to an individual's needs. Fundamentally the distinctive and evolving network of relationships in which young people are embedded must be revealed and taken into account if the support provided to them by professionals is to be most effectively tailored to their needs.

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ORIGINAL ARTICLE

Being as Normal as Possible: How Young People Ages 16–25 Years Evaluate the Risks and Benefits of Treatment for Inflammatory Arthritis

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Objective. **To explore how young people (ages 16 25 years) with inflammatory arthritis evaluate the risks and benefits of treatment, particularly treatment with biologic therapies.**

Methods. **This qualitative study involved in-depth interviews (n = 44) with young people, trusted others (e.g., parents), and health professionals; audio-recordings (n = 4) of biologic therapy related consultations; and focus groups (n = 4). Analysis used techniques from grounded theory (open and focused coding, constant comparison, memoing, and mapping).**

Results. **Young people aspired to live what they perceived as a "normal" life. They saw treatment as presenting both an opportunity for and a threat to achieving this. Treatment changes were therefore subject to complex and ongoing evaluation, covering administration, associated restrictions, anticipated effects, and side effects. Information sources included expert opinion (of professionals and other patients) and personal experience. Previous treatments provided important reference points. Faced with uncertain outcomes, young people made provisional decisions. Both trusted others and health professionals expressed concern that young people were too focused on short-term outcomes.**

Conclusion. **Young people value treatment that helps them to live a "normal" life. There is more to this than controlling disease. The emotional, social, and vocational consequences of treatment can be profound and lasting: opportunities to discuss the effects of treatment should be provided early and regularly. While making every effort to ensure understanding of the long-term clinical consequences of taking or not taking medication, the wider impact of treatment should not be dismissed. Only through understanding young people's values, preferences, and concerns can a sustainable balance between disease control and treatment burden be achieved.**

INTRODUCTION

There is increasing consensus that patient preferences are both important and unpredictable. What patients want is not necessarily what doctors think they want (1). Within rheumatology, the body of studies examining patient treatment preferences is growing. However, research to date has focused on older adults with rheumatoid arthritis (2–5).

Like older adults, young people with inflammatory arthritis (IA) can have severe disease warranting aggressive treatment, including biologic agents (6). In other respects, however, they are different. Clinical differences set young

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Significance & Innovations

- Prior research on patient treatment preferences has focused on older adults with rheumatoid arthritis. Young people with inflammatory arthritis have different circumstances and concerns.
- Young people see treatment as presenting both an opportunity and a threat to their desire to lead a normal life. They describe a wide range of consequences (physical, emotional, social, and vocational) arising from their treatment regimens.
- In evaluating treatment changes, young people take this wide range of outcomes into account; where outcomes are uncertain they consider decisions to be provisional.
- Young people need active encouragement to discuss their treatment concerns and difficulties with the care team, so that a sustainable balance between disease control and treatment burden can be achieved.

people with juvenile idiopathic arthritis (JIA) apart from adult patients. Developmental differences further distinguish them (and young people with other forms of IA) from adult patients, who bring fully matured brains to bear on their decision-making (7). The social context in which young people make decisions about managing their disease also differs in important ways from that of older patients, and may have a profound effect on decision-making (8).

Choice is exercised within and outside the clinic. Patients make decisions about treatment options in the context of consultations, and then, on a routine basis, whether and how to enact their agreed upon regimen. Evidence of the link between patient preferences and adherence is increasingly convincing: where treatment decisions align with patient preferences, clinical outcomes are better (1,9,10). Treatment choices may also promote (or impede) the achievement of key developmental milestones, such as establishing a career and a family. So the consequences of treatment decisions made early in life may affect both short- and longer-term health, intrude into other domains, and extend through the life course.

There are a variety of reasons that young people's treatment preferences might differ from those of older adults. Understanding how they inform treatment choices matters, due to the profound and lasting impact that such decisions have. Our study therefore explored how young people evaluate the risks and benefits of treatments, in particular biologic therapies. It considered their priorities and concerns and the challenges treatment presented. Other aspects of the work (relating to the influence of "trusted others" on decision-making) have been reported elsewhere (11).

PATIENTS AND METHODS

We report findings from a qualitative study conducted in England, 2012–2014. The study explored decision-making about biologic therapies by young people, ages 16–25 years, with a diagnosis of IA (JIA, ankylosing spondylitis [AS], psoriatic arthritis [PsA], or rheumatoid arthritis [RA]). Subject to meeting nationally agreed upon criteria, young people in England can access a range of treatments, including biologic therapies, without charge, from the National Health Service (NHS).

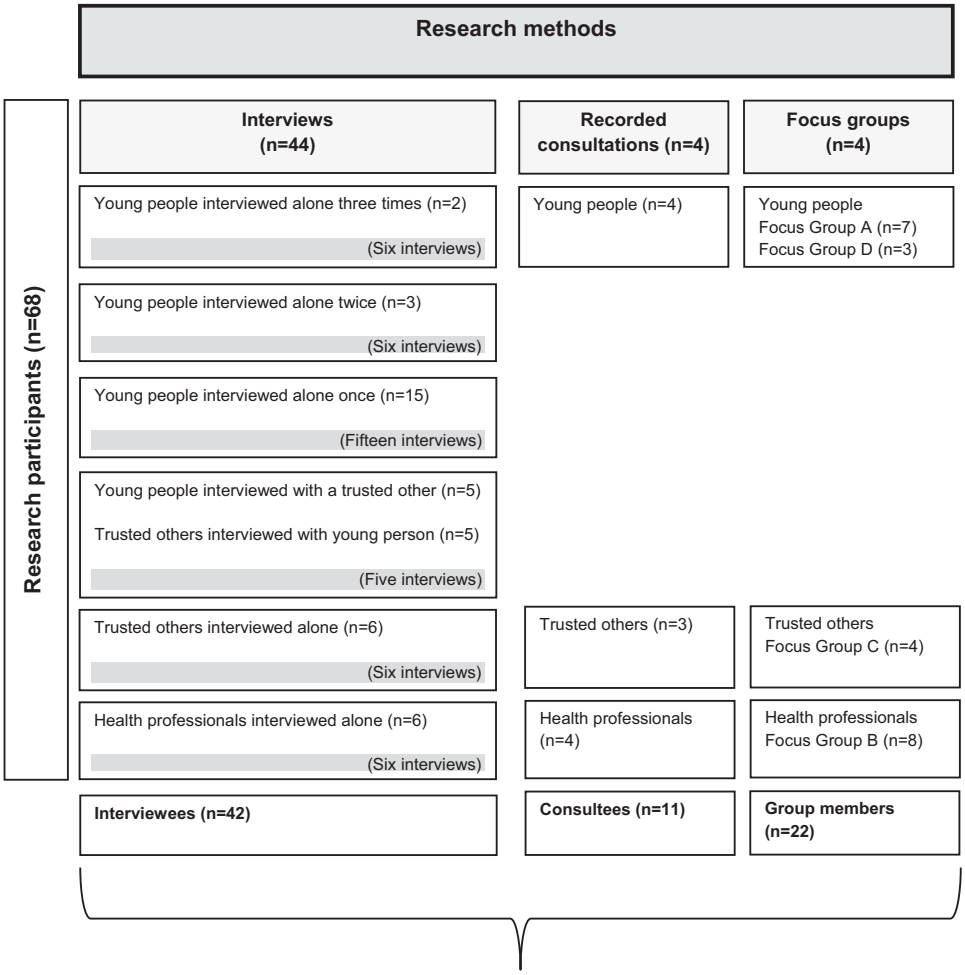
The study used multiple methods and sources, an approach termed methodological and data triangulation (12). Methods used included interviews (n 44), audio-recordings of consultations (n 4), and focus groups (n 4). Research participants (n 68) were young people (n 37), trusted others (n 15), and health professionals (n 16). Figure 1 maps methods against participants. Participants were recruited via 3 NHS Hospital Trusts running adolescent, young adult, and/or adult rheumatology clinics. All participants consented verbally and in writing and the study had research ethics committee approval (Yorkshire & Humber, Leeds East). The research complied with the Declaration of Helsinki.

Interviews. Twenty-five young people were interviewed, recruited purposively to ensure diversity (in demographic characteristics, diagnosis, and treatment history) and support exploration of emerging issues (see Table 1). The sample included 5 young people who at the first interview had not yet been offered a biologic agent, 5 who had recently been offered a biologic agent, and 15 taking (or who until recently had taken) a biologic agent. Where treatment status changed, we sought to reinterview (3 young people were interviewed twice, and 2 others were interviewed 3 times). Young people were recruited with the help of the direct care team, which identified patients with specified characteristics and made initial approaches (giving young people written information and seeking permission to pass on their contact details).

Eleven trusted others also took part in an interview (8 mothers, 1 father, 1 grandmother, and 1 partner). Trusted others were identified by the participating young people. In 5 instances they were interviewed with the young person. Trusted others who did not accompany young people to interviews were approached through them. Young people gave trusted others written information on the study and asked for permission to pass on their contact details. Most agreed to participate; those declining included a close friend and a partner.

Six health professionals were interviewed, including nursing and medical staff from all 3 types of clinic and trusts, recruited to provide a range of perspectives. Interviewees were proposed by the research team and approached directly by the researcher with written information.

Interviews lasted 40–120 minutes. Most were conducted face-to-face, at a location of the interviewee's choice. All interviews were semistructured, using schedules informed by the team's experience, a review of the literature, and the emerging analysis. These addressed a set of core topics (e.g., the decision-making process, information exchange, views on risks and benefits) but differed in detail and emphasis to reflect individual circumstances (e.g., young people's treatment history and the specifics of professionals' roles).



Note: These numbers (42+11+22) sum to more than 68 as six research participants provided data via more than one research method (5/6 providing data via two methods and 1/6 providing data through three methods).

Figure 1. Map of research methods and research participants.

Recorded consultations. Four consultations were recorded involving 11 participants (different combinations of young people, trusted others, and health professionals). We hoped to compile a larger body of recordings, but negotiating and arranging these proved challenging. The recordings should be considered a convenience sample. However, they include a short first conversation about biologic therapies and a lengthier counseling session. They provide a detailed record of how biologic therapies are explained and the questions and concerns that arise.

Focus groups. Four focus groups were convened at the end of the project to explore the face validity of findings. Focus group A comprised 7 young people (5 female, 2 male; ages 16–20 years; 5 with JIA, 2 with other diagnoses) and focus group D comprised 3 young people (all male; ages 17–22 years; 2 with JIA, 1 with AS). Focus group B comprised 8 health professionals with interests in adolescent rheumatology. Focus group C comprised 4 trusted others (3 mothers, 1 grandmother). Recruitment to the focus groups was purposive (invitations being extended to people with and without prior involvement in the research). The groups provided a forum in which participants could comment on the intelligibility, credibility, and significance of findings and invite further reflection on the analyses.

Data analysis. Interviews, focus groups, and consultations were audio-recorded and transcribed verbatim for analysis. Transcripts were analyzed by an author (RIH) using techniques foundational to grounded theory (open and focused coding, constant comparison, memoing, mapping) to identify patterns and relationships (13,14). Data were analyzed within and across samples. The principal investigator (TR) provided a check by analyzing data segments (selected transcripts or data on a particular theme). Analyses were tested further with other researchers in biweekly data clinics and at biannual team and steering group meetings. Analysis ran alongside and informed sampling.

RESULTS

We report here on a series of related themes in our data, all underpinned by the concept of "being normal." This

Table 1. Characteristics of young people interviewed (n = 25)

Characteristic	No.
Diagnosis	
Juvenile idiopathic arthritis	15
Ankylosing spondylitis	7
Psoriatic arthritis	2
Rheumatoid arthritis	1
Female	15
Mean (range) age, years	20 (16 25)
Ethnicity	
White British	24
Mixed	1
Mean (range) disease duration, years	9 (<1 to >20)
Type of medication taken at time of final interview	
Biologic agents	19
Disease modifying antirheumatic drugs	8
Oral steroids	1
Nonsteroidal antiinflammatory drugs	8
No medication	3
Rheumatology service accessed	
Adult clinic	10
Young adult clinic	8
Adolescent clinic	7

was a high priority for young people in our study, informing the processes of making and evaluating decisions. Put simply, young people aspired to live a normal life (theme 1); treatment was perceived as both an opportunity for and a threat to this (theme 2). Powerful emotions were a context for and a consequence of treatment decisions (theme 3). Information relevant to such decisions was acquired from different sources (theme 4), which did not always align. Decisions were considered provisional (theme 5) and were reviewed against experience. The focus on short-term outcomes (theme 6) was a concern to trusted others and professionals.

Theme 1. Aspiring to live a normal life. Many of the young people interviewed talked of how they aspired to live what they perceived as a normal life, or as one put it, "to get back to . . . a normal way of life . . . not really too different from anybody else's" (male, age 21, AS). Not all the young people taking part in the focus groups connected with the term "normal," but they accepted that its components had resonance: "Living a normal life is a priority? I would agree with [that], but don't like the word 'normal"' (male, age 24, AS).

"Normal" is a complex, multifaceted, situational, and dynamic concept concerning not only bodily function and experience, but also mental well-being and performance of social and vocational roles. One professional explained: "They want to be able to get up in the morning and just be able to move. They want to go to work. They want to stay at college. They want to complete their university degree. They want to travel. They want to do normal things" (health professional).

Young people constructed their ideas of normal through reference to alternate selves (pre-diagnosis or on a good day) and to others. They engaged in processes of implicit and explicit comparison, with unknown, idealized others (young people in the abstract) and known others (e.g., siblings and friends). These known others seemed a particularly important reference point. Young people wanted to feel, look, think, and act like them. They largely wanted their peers to think they were like them, and to treat them accordingly. For some young people this hope was realized, but for others, the sense of being different (a word commonly juxtaposed with normal) ran deep.

Theme 2. Seeing treatment as an opportunity and as a threat. In the context of aspiring to live a so-called normal life, treatment appeared to be both an opportunity and a threat. Most young people confronted with treatment decisions were keen to experience relief from symptoms. A change in treatment might reduce pain, improve mobility, and get life "back to normal." Effective management of their condition enabled young people to make plans for the future. This included taking steps into education or employment, or toward independent living: "I'm thinking of going back to college in September. Just because I feel like everything is being managed now. I feel I've a better chance of going to college and actually being able to stay there" (female, age 24, PsA).

However, those experiencing side effects from prior treatments were acutely aware of the potential for less positive outcomes, presenting threats to normal life. Some young people (and trusted others) described how steroids had caused changes in face shape and weight. Many talked of how methotrexate had triggered nausea, vomiting, and vocational underperformance: "Every Monday, you could guarantee that I wasn't at school . . . 'cause I was still sick from the medicine" (female, age 17, JIA). Others explained how increased susceptibility to infections had diminished their general sense of well-being and disrupted their lives.

Even where a drug proved effective in controlling inflammatory processes, relieving associated symptoms, and had minimal side effects, it could be highly intrusive. Several young people explained how their plans and routines were dictated by their treatment schedule. The loss of freedom to engage in activities taken for granted by their peers was lamented, compounding a sense of being different: "All my friends are going out, going clubbing, going camping. And I always have to think about my medication and that first, before I even think about anything else" (female, age 22, JIA).

Theme 3. Experiencing powerful emotions. Many young people revealed anxieties about aspects of treatment. Methotrexate cast a long shadow over some young lives: "The look of it, the smell of it, the very thought of it made me shake" (male, age 17, JIA). The psychosocial sequelae of treatment included familial tensions, isolation, and bullying. Decisions about treatment changes were consequently highly charged: seemingly small adjustments to dosage, routines, or mode of administration could unleash strong emotions. Escalation of treatment forced young people to confront their condition, challenging efforts to perceive and present themselves as normal. Such decisions also brought them face-to-face with the uncertainties of the future. Most recognized that

no treatment was guaranteed to work, and that their options were becoming more limited. Young people tried to maintain an optimistic outlook, but past disappointments bred caution: "The methotrexate worked for a little bit and then it stopped working … to put all your hopes on (adalimumab) seems a bit … I can't really do that" (female, age 24, PsA).

Theme 4. Acquiring information from different sources. In evaluating treatment changes, young people drew on information from various sources. They gained much of their knowledge, particularly about biologic agents, from health professionals delivering their care. However, this was supplemented by information from family or friends with relevant experience or expertise, research done by and for young people, and, critically, direct personal experience.

Evidence from personal experience played a powerful role in shaping young people's understanding of their condition and the treatments used to manage it. Prior treatments, including for other conditions, provided important reference points: "My attitudes toward potential outcomes with things like this are colored by a lot of the treatments I've received as a child" (male, age 22, AS). Often experiential evidence aligned with clinical measures, but not always: "Sometimes I feel like I'm just a blood results number. They, they're looking at my blood results—yes, my blood result may be sky high, but I feel perfectly fine" (female, age 20, JIA). Conflicting evidence could lead to frustration (on all sides) and fueled a sense of uncertainty.

Theme 5. Making provisional decisions. Young people emphasized the uncertainty associated with a new treatment, with respect to both its effects and challenges: "Nobody knows which one's best (etanercept or adalimumab) … it's a bit of a shot in the dark" (male, age 25, AS). New treatments were judged against other (past or present) treatments and, less frequently, the uncontrolled condition. Often the push of a certain (and intolerable) past or present treatment outweighed the pull of a future treatment: "I was willing to try it (etanercept) because I hated methotrexate" (female, age 16, JIA).

In the face of uncertainty, treatment decisions were considered provisional and open to review. Having gathered information on treatment administration, associated restrictions, anticipated effects, and potential side effects, the approach most often adopted was "try it and see." Ultimately the test was whether treatment made life "easier … rather than harder" (female, age 22, JIA). This required more than just an improvement in symptoms; also important were side effects that were at most "annoying," minimal restrictions, and a relatively simple and stable regimen. Collectively, these things acted to increase or diminish the sense of living a "normal" life. Where life did not feel more normal, commitment to treatment waned. Commitment could be reinforced by experiential evidence acquired from suspension of treatment: "When I came off medication, and I'd flare a bit or something, I realized how much difference it's doing … that has made me understand how they are, how it is doing me good … although sometimes, some days, it feels like it's making me worse" (male, age 16, JIA). Both young people and trusted others described trialing withdrawal of treatment; other reasons for breaks included infection, surgery, travel, conception, and oversight.

Where the impact of treatment was rapid and clear cut, as was often the case with biologic agents, the "try it and see" approach was unproblematic. However, with drugs such as methotrexate, where benefits took longer to emerge and initial side effects could be onerous, decisions were often reconsidered. Some young people (or trusted others) consulted care teams about alternatives. However, not all were aware that there were alternatives, and could wait until their next appointment to discuss them. In such circumstances the potential for unilateral discontinuation or partial nonadherence to treatment was high, with care teams becoming fully aware of young people's difficulties only when a crisis point was reached.

Theme 6. Focusing on the short term. Trusted others and health professionals perceived young people as focused on short-term outcomes: "My worry is always years ahead, where [my daughter] wouldn't worry about [the future] at all. That will not even feature in her, in her mind" (mother of female, age 17, JIA). They expressed concern that young people might not take on the longer-term risks of, on the one hand, taking treatment and, on the other, not taking treatment: "You're giving [treatment] to them to help them live a normal life. But there's much more to it than that. You give it to them … to stop things happening that they couldn't even begin to imagine" (health professional).

Short-term concerns are more prominent in the data from young people than longer-term concerns. The minority vocalizing longer-term concerns had more complex treatment histories or a history of cancer in the family. More commonly, young people noted the long-term risks briefly, framing them as low probability and easily resolved. A common assertion was that doctors would not propose treatment options if the risks were unreasonable: "The view I take is that if it's been offered to me, it's safe" (male, age 25, AS). However, a small minority reflected that when beginning certain treatments, they had not appreciated that they might be taking them long term.

Lack of attention to the long-term risks was for some young people an active choice. Many stressed how difficult it was to think beyond the immediate future when life was made so challenging by their condition and/or treatment. They felt bound to accept the long-term uncertainties or, as one interviewee put it, the "what ifs," in order to get on with life in the short term. They acknowledged, but tried not to dwell upon, the future effects of treatment: "If something bad happens, I'll cross that bridge when I come to it. For now, it's just keeping me normal. I know that sounds a bit reckless, and I don't mean it like that, but like, I can't worry about what, you know, what would happen" (female, age 24, PsA).

DISCUSSION

Despite the growing interest in patient preferences, to date little attention has been given to those of young people

with IA. Lipstein et al (15) describe treatment decision-making by adolescents with chronic illness generally as an understudied area. Their work, which includes young people with JIA, focuses on early adolescence (ages 10–15 years). Our research on older adolescence and early adulthood (ages 16–25 years) extends and complements that. It provides new insights on the perspectives of an important but neglected patient subgroup, and on how young people's experiences are understood by those around them (trusted others and health professionals).

Our study shows clearly how young people with arthritis, similarly to those with other chronic conditions (16,17) aspire to live so-called normal lives, but that both IA and its treatment present challenges to this. We recognize that young people are not alone in valuing treatments promoting "normal" life: studies of older adults with arthritis have drawn similar conclusions (3,18). However, our research suggests that the features of normal life in adolescence and early adulthood are distinct, as are the consequences of not being normal or not being perceived as normal. It shows how participation in developmental and peer-group activities acts as a litmus test for normality, and constraints upon participation may negatively affect well-being, careers, and relationships. Williams et al note how a socially derived concept of normality becomes more prominent in adolescence (17). In illuminating how this informs young people's treatment preferences, our work reinforces and extends the findings of other research on chronic illness in adolescence (19–21).

Treatment developments in rheumatology have challenged thinking about the disruptive nature of chronic illness (22,23). New medications have been framed as restorative by some authors (24). However, treatments such as methotrexate are widely recognized as having unpleasant side effects (25). Beyond rheumatology, increasing attention is being given to the practical and cognitive burdens that treatment regimens place on patients (26). Our study reflects this complex picture, showing how for young people with arthritis, treatment is a double-edged sword, and adding to evidence suggesting that clinical outcome measures do not capture all that matters to patients (3,27,28).

Our research highlights the preferences of a diverse group of young patients, but is no substitute for careful exploration of the concerns of individuals. Other authors have stressed the importance of dialogue with older patients before and after the initiation of treatment (5). This is no less the case for young people; indeed they may need more active encouragement, and a wider range of channels, to raise concerns and articulate difficulties. A central message from our work is that it must be made absolutely clear to young people that concerns about the impact of treatment on, among other things, appearance, relationships, or education, are valid things to raise. A recent study has shown that bidirectional sharing of information in pediatric consultations about biologic agents is uncommon (29). Clearly, a more collaborative approach to considering, constructing, and sharing preferences is needed. Models such as shared decision-making and collaborative deliberation offer relevant processes and guidance (30,31).

A key concern of trusted others and health professionals was young people's perceived focus on the short term. Care must be taken not to discount prioritization of short-term gains as a reflection of the adolescent brain. While neuroscience offers plausible explanations for such a short-term perspective, social context has been shown as important (8). Moreover, older adults with arthritis have been found to have a similar focus (2,18,28). This is not to dispute the importance of making sure that young people are well informed about risk-benefit tradeoffs or longer-term consequences of not taking clinically optimal medication (or not taking medication the optimal way) (15). Young people have themselves reported wanting transitional care programs to cover rationales for treatment, side effects, and delays in observation of benefit (32). We believe that there is a strong case for periodically revisiting the long-term risks and benefits of treatment decisions (to take or not), both as a prompt to young people to air concerns and to check understanding.

Our research has both strengths and weaknesses. The nature of qualitative work is that it is in-depth but small scale, with a consequent strength being the richness of data and a weakness being the low number of cases. However, the body of data compiled during this study is relatively substantial and the number of participants (n 68) relatively large. Conducting serial and triangulated interviews enabled us to confirm stories and capture evolving perspectives (33). Recording consultations enabled us to compare what people do against what they say they do in these contexts. The use of focus groups to explore the face validity of our findings (also referred to as member validation) strengthens our confidence in their credibility (34).

In conclusion, this study provides important insights into young people's circumstances and preferences and how these may inform treatment decisions. It challenges assumptions, implicit in much of the previous research, that studies of the typical (i.e., older) patient provide a basis for understanding young people's priorities. Our data reveal age as an important factor in the evaluation of treatment options, underpinning perceptions of "normal" life, issues achieving and maintaining this, and the consequences of not doing so. But to find out what matters to any particular young person, we must ask questions, set aside our assumptions, and listen to what he or she has to say. Only through understanding young people's values, preferences, and concerns can a sustainable balance between disease control and treatment burden be achieved.

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AUTHOR CONTRIBUTIONS

All authors were involved in drafting the article or revising it critically for important intellectual content, and all authors

approved the final version to be submitted for publication. Ms Hart had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study conception and design. McDonagh, Thompson, Foster, Kay, Rapley.

Acquisition of data. Hart, McDonagh, Thompson, Foster, Kay, Myers.

Analysis and interpretation of data. Hart, Rapley.

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WILEY

RESEARCH ARTICLE

What impact does written information about fatigue have on patients with autoimmune rheumatic diseases? Findings from a qualitative study

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Abstract

Objectives Although fatigue is a common symptom for people with rheumatic diseases, limited support is available. This study explored the impact of written information about fatigue, focusing on a booklet, *Fatigue and arthritis*.

Methods Thirteen patients with rheumatic disease and fatigue were recruited purposively from a rheumatology outpatient service. They were interviewed before and after receiving the fatigue booklet. Two patients, plus six professionals with relevant interests, participated in a focus group. Transcripts were analysed thematically and a descriptive summary was produced.

Results Interviewees consistently reported that fatigue made life more challenging, and none had previously received any support to manage it. Reflecting on the booklet, most said that it had made a difference to how they thought about fatigue, and that this had been valuable. Around half also said that it had affected, or would affect, how they managed fatigue. No one reported any impact on fatigue itself. Comments from interviewees and focus group members alike suggested that the research process may have contributed to the changes in thought and behaviour reported. Its key contributions appear to have been: clarifying the booklet's relevance; prompting reflection on current management; and introducing accountability.

Conclusions This study indicated that written information can make a difference to how people think about fatigue and may also prompt behaviour change. However, context appeared to be important: it seems likely that the research process played a part and that the impact of the booklet may have been less if read in isolation. Aspects of the research appearing to facilitate impact could be integrated into routine care, providing a pragmatic (relatively low-cost) response to an unmet need.

KEYWORDS

fatigue, rheumatic diseases, written information, qualitative research

1 | INTRODUCTION

As previously documented in *Musculoskeletal Care* (Farren, Goodacre, & Stigant, 2012) and elsewhere (Hewlett, Cockshott, Byron, Kitchen, & Tipler, 2005; Mengshoel, Norheim, & Omdal, 2014; Overman, Kool, Da Silva, & Geenen, 2016; Schoofs, 2001), fatigue is a significant and burdensome symptom for people with autoimmune rheumatic diseases. It appears to reduce health-related quality of life substantially and may in some instances have a greater impact than the more widely attended to symptom of pain (Kirwan & Hewlett, 2007). However,

despite the prevalence and impact of fatigue, effective care strategies are yet to be established and clinicians often struggle to address it when raised (Repping-Wutts, van Riel, & van Achterberg, 2008). Although there is evidence to suggest a biological as well as a psychosocial basis for fatigue (Newton & Jones, 2010), pharmacological therapies appear to have limited effect, even where they have proved effective for pain and inflammation (Chauffier, Salliot, Berenbaum, & Sellam, 2012; Ng & Bowman, 2010).

Non-pharmacological strategies have been found to benefit some patients with fatigue and other long-term conditions (Patterson, Wan, & Sidani, 2013). A systematic review (Cramp et al., 2013) found some evidence of benefit for psychosocial interventions and physical activity in managing fatigue linked to rheumatoid arthritis (RA). Group programmes, delivered by clinical psychologists and underpinned by cognitive behavioural therapy (CBT), have been judged as showing particular promise (Hewlett et al., 2011); research is under way to explore whether other rheumatology professionals might deliver such programmes to similar effect (Hewlett et al., 2015). However, although promising, such programmes are unlikely ever to be available, accessible and attractive to all patients in need of education and support (Thompson, 2011).

Other patients may turn, or be directed, to self-management resources in the shape of written information in either print or electronic form. These materials have featured as the 'usual care' arm of trials of group programmes (Hewlett et al., 2011, 2015). However, despite appearing to offer a pragmatic solution to the information and support needs of patients unable or unwilling to access group programmes, there is little evidence that such materials are a widespread and consistent feature of usual care.

The use of patient information materials in trials (Hewlett et al., 2011, 2015) is providing useful outcome data in the form of clinical and other measures. However, information on the processes involved – how patients perceive, interact with and ultimately employ such resources – remains limited. With the exception of the early work of Bishop, Barlow, Williams, and Hartley (1997), patient literature (in rheumatology) has had surprisingly limited scrutiny in its own right. The present study set out to fill these potentially important knowledge gaps, by exploring patients' response to the Arthritis Research UK publication *Fatigue and arthritis* (Arthritis Research UK, 2011). This is a 24-page booklet describing the features and causes of fatigue, and recommending a range of strategies to reduce its impact. The advice contained is broadly consistent with that provided online by the other organizations producing information for people with autoimmune rheumatic diseases (e.g. the US-focused Arthritis Foundation). However, the booklet offers greater detail, is available as a hard copy and includes practical tools such as a chart for monitoring activity and fatigue.

2 | METHODS

This paper reports the findings of a study, conducted in England over the period 2014–2015, investigating the reception, use and impact of the *Fatigue and arthritis* booklet by and on patients using a rheumatology outpatient service. The methodological approach taken was qualitative description, as described by Sandelowski (2000, 2010). This is a pragmatic, naturalistic approach to qualitative research, which focuses on producing low-inference descriptions of experiences and events. It is particularly suited to producing 'minimally theorized' findings (Sandelowski, 2000) of practical value to practitioners and policymakers. As such, it fitted well with our ambitions for the project.

2.1 | Sampling and recruitment

Qualitative description favours the construction of a non-random sample reflecting the diversity of a given population (a goal often referred to as achieving 'maximum variation'). Samples need to be of an adequate size to support this. Based on prior experience of treating and researching this patient group (Hart et al., 2015; Lee, Thompson, Whybrow, & Rapley, 2016; Thompson, 2011), the team predicted that a sample of 12–15 patients would be sufficient to accommodate potentially significant areas of variation and to achieve 'data saturation' (where no new themes, ideas or issues emerge). Ultimately, 13 patients were recruited for interview over a period of approximately 12 months. Two patients, one of whom had taken part in interviews, were recruited to the 'expert' focus group convened at the end of the study.

The sample was constructed purposively, with ongoing attention to diagnosis, gender, age and fatigue severity (see Table 1), as well as wider health, including mental health; social, occupational and domestic backgrounds; and life demands. Our concern was to ensure variety within the sample, so while fatigue severity was assessed using the Fatigue Impact Scale (FIS) (Fisk et al., 1994), no particular level of fatigue was set, a priori, as a condition for in−/exclusion. Instead, potential participants were eligible if they had been diagnosed with one of the inflammatory rheumatic diseases specified (ankylosing spondylitis, primary Sjögren's syndrome or rheumatoid arthritis), reported fatigue which they felt was significant, and their fatigue was judged to be related to the rheumatic disease and not to another condition (e.g. anaemia, hypothyroidism). Similarly, participants were asked to complete the Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983) to enable us to assess variation in, and characterize more fully, the wider health of our sample.

Potential research participants were identified by clinical members of the research team during routine clinical encounters on the basis of their professional knowledge and with reference to patient records. As sampling progressed, and patients with more specific characteristics were sought, help was sought from other clinicians working in the rheumatology outpatient service. Initial approaches to prospective participants were made by clinicians, with expressions of interest subsequently followed up by the project researcher (the first author).

Six health professionals (with interests in musculoskeletal care, fatigue and/or patient education) were recruited to the study as focus group participants. The approach was again purposive and the group included two clinicians (from within and outwith rheumatology), a nurse and three allied health professionals (from occupational therapy, psychology and physiotherapy services). Professional participants were identified by the research team but approached directly by the project researcher. All potential participants (patients and professionals) were given written information on the study and in turn provided written consent.

TABLE 1 Characteristics of interviewees (n = 13)

Interviewee	Diagnosis[a]	Gender	Age (years)	FIS score[b]	HADS scores[c] (A, D)
A	pSS	Female	77	63	4,4
B	pSS	Female	70	56	6,5
C	AS	Female	52	71	5,9
D	RA	Male	25	45	6,5
E	AS	Male	32	88	13,7
F	RA	Female	29	33	6,4
G	RA	Female	40	36	8,8
H	AS	Male	58	74	7,7
I	pSS	Female	70	61	5,9
J	pSS	Female	63	56	5,3
K	RA	Female	65	97	9,10
L	pSS	Male	61	99	11,8
M	AS	Female	57	78	8,3

[a]One of three specified inflammatory rheumatic diseases: primary Sjögren's syndrome (pSS); ankylosing spondylitis (AS); and rheumatoid arthritis (RA).

[b]The Fatigue Impact Scale (FIS) is a 40-item questionnaire exploring three dimensions of fatigue. Scores may range from 0 to 160, with higher scores indicating higher fatigue impact.

[c]The Hospital Anxiety and Depression Scale (HADS) is a 14-item questionnaire with two subscales [Anxiety (A) and Depression (D)] each running from 0 to 21. Scores of 0 7 are within the 'normal' range. Scores of 11 and above indicate a probable mood disorder.

2.2 | Data collection

Data were collected through serial semi-structured interviews (26) and a focus group (one). These methods support exploration of 'complex phenomena' (Tong, Sainsbury, & Craig, 2007) with interviews, in particular serial interviews, generating rich, contextualized data on individual experiences, beliefs and values (Lee et al., 2016; Murray et al., 2009; Ong & Richardson, 2006; Paskins & Hassell, 2012). Focus groups give access to different forms of expression, and perspectives arising as a consequence of social interaction (Kitzinger, 1995). The use of more than one method, or 'methodological triangulation', enriches understanding and supports validation (Denzin, 1989).

Patients were interviewed by the first author (a social scientist with prior experience of qualitative research in rheumatology) before and after being given a copy of the *Fatigue and arthritis* booklet. Interviews took place in settings chosen by interviewees (their homes, the hospital, university and a café). Initial ("pre-booklet") interviews explored patients' circumstances and their experiences of, and efforts to manage, fatigue. Follow-up ("post-booklet") interviews typically took place around four to six weeks later and explored patients' impressions and use of the fatigue booklet, and the impact they perceived it having. Interview guides were drafted at the start of the study by the project researcher, with input from the wider team and patient partner. They were revised as the study progressed to take account of preliminary analyses and in response to the statements of each interviewee. It is more appropriate to view them as guides for conversation than as prescriptive scripts. The interviews, which lasted from 23 to 132 minutes, were all recorded and transcribed verbatim.

The focus group was convened in the final phase of the project in order firstly to explore whether interaction between patients and professionals might draw out alternative perspectives on the booklet (contrasting with each other or with those emerging in interview). Its secondary function was to invite critical reflection on the findings from the interviews and their potential implications for practice. All participants were sent a copy of the booklet ahead of the event and asked to read it by way of preparation. Topics explored include participants' views on the booklet, perspectives on the key findings from the interviews, and thoughts as to the future development and use of fatigue-related educational resources. The focus group was facilitated by the first author and took place in a hospital meeting room. It ran for 90 minutes and was recorded and transcribed.

2.3 | Data analysis

Data analysis ran alongside and informed data collection. Transcripts were checked for accuracy and then systematically analysed. The initial analysis, undertaken by the first author, involved line-by-line coding (Charmaz, 2006) to identify and abstract salient features of individual transcripts. Working within broad a priori themes (which also informed data collection and reporting), data and codes were then compared, sorted, related and (in the case of some codes) combined, until patterns, exceptions and revealing illustrations could be identified. Case summaries, charts, diagrams and memos were employed both to facilitate the process of analysis and to provide the wider team with access to the data and the analytical logic. Meetings with the wider team, which included a patient partner, and an external expert, encouraged reflexivity and improved analytical rigour.

2.4 | Ethical approval

The study had Research Ethics Committee approval from the Proportionate Review Sub-committee, National Research Ethics Service Committee Yorkshire & Humber-Leeds East (ref. 14/YH/1054). It complied fully with the Declaration of Helsinki.

3 | RESULTS

We begin this section by outlining how interviewees were affected by and tried to manage fatigue, before receiving the booklet. We then detail their responses to the booklet, and the changes in thought and behaviour they reported. We close by considering how the research experience itself may have contributed to those impacts. Data from the expert focus group are incorporated where they illuminate a point or provide an alternative perspective.

3.1 | Experience and management of fatigue prior to receipt of the booklet

Our interviewees had varying histories of fatigue and rheumatic disease. For some, these were longstanding problems; for others, they were more recent developments. Although describing different patterns of fatigue, interviewees consistently reported that it made life more challenging and less rewarding. Fatigue disrupted activities and increased their physical and/or mental demands. Motivation to engage in social or leisure activities was undermined:

> *Socializing, just doing things that you want to do, are rather harder, or get put on hold, because you're tired all the time. [Interviewee D]*

Often, people did not understand their fatigue, or connect it with their condition:

> *It hadn't occurred to me that it might be part and parcel of the condition. [Interviewee A]*

Instead they attributed it to age, apathy or other – undiagnosed – illnesses. This lack of understanding left them feeling guilty and anxious about their work, domestic and social lives:

> *You feel like you're lazy, you know. I sort of come in and I'm thinking, you know, "Eeh, I'm such a lazy so-and-so". [Interviewee C]*

People worried about how their difficulties were perceived and judged by others, and the additional challenges the future might bring.

Interviewees described having made changes to their lives, to try to deal with fatigue. These included: conserving energy; managing demands by planning ahead; taking breaks for rest and recovery; and looking after themselves better. Some of these changes were active and pre-emptive choices but others – such as rest – were often reactive – that is, precipitated by overwhelming fatigue. What emerged strongly from the data was that, even where people had identified helpful strategies, they struggled to use these consistently:

> *Some days, I handle it really badly… I won't pace myself, some days I, I still, just approach things badly, or just won't talk to people'. [Interviewee E]*

No one reported having professional support to identify or implement fatigue management strategies.

Overall, fatigue-related communication with health professionals (in primary or specialist care) appeared limited; for some, the initial interview was the first time they had talked about fatigue at any length. In addition to general difficulties with regard to communication in medical consultations, the data suggest a number of barriers specific to fatigue. These include: reliance on a colloquial vocabulary ("so tired", "exhausted", "knackered", "wiped out", "done in"); uncertainty about fatigue's relationship to rheumatic disease; doubt as to fatigue's 'place' on the consultation agenda; and a belief that nothing can be done. These barriers affected if and how concerns were shared, and could be reinforced by clinicians' reactions to disclosures of fatigue. Patients wanted professionals to initiate regular discussions:

> *It would be great if the consultants did say to you "And how are your fatigue levels?" But that doesn't happen. It doesn't happen. [Focus group participant (FGP) T]*

3.2 | Response to the booklet: Reported changes in thought and behaviour

When asked what impact the booklet had upon them, interviewees typically reported that it had made a difference to how they thought about fatigue, and that this was of real value. Understanding fatigue and its association with rheumatic disease helped to allay fears that fatigue was a sign of another, undiagnosed health problem or an inevitable age-related decline:

> *The relief… of recognizing that it's part of the condition, not that I was sinking into an age-related depression. [Interviewee A]*

It validated interviewees' experiences and concerns, and somewhat alleviated the guilt associated with decreased activity:

> *'Makes you feel a bit more like you're not making it up. [Interviewee F]*

The booklet gave interviewees access to new ways of defining and describing their experience, enabling and encouraging the discussion of fatigue and its impacts:

> *I think I just need to be more honest, I suppose, and not try to cover up [Interviewee M]*

Critically, it conveyed the message that it was possible to target important drivers of fatigue and, by doing so, reduce its impact:

> *There's things out there you can … incorporate into your life to make you feel better. [Interviewee G]*

This sense that things could be done was a starting point, and a powerful motivator for change.

Interviewees largely valued the prompt to reflect on their current practice (i.e. efforts to maintain routines despite fatigue, or to manage fatigue):

> *It is useful now and again just to … think about it and maybe analyse it, analyse what you're doing, and if there is any, any changes that you can effect, because you tend just to go on with the same thing. [Interviewee H]*

Around half said that the booklet had affected, or would affect, their approach to managing fatigue, and reported making, or planning to make, small but potentially significant adjustments to their behaviour. This included adopting practices broadly in line with 'the four Ps' (pacing, prioritizing, planning and problem-solving (Arthritis Research UK, 2011)). Behaviours aligned with 'pacing' and 'prioritizing' featured in a particularly wide range of accounts, with interviewees describing new patterns of rest and activity, novel strategies for conserving energy and efforts to review and more vigorously prioritize commitments:

> *Just looking at what I do in a day … just trying to decide, really, "Yes, that needs to be done. That can wait. And that, it doesn't really matter if I don't do (it)". [Interviewee G]*

Several interviewees intended to monitor their energy output more closely and some had begun to schedule pleasurable as well as utilitarian activities:

> *We're just trying to like, go out and do things, trying to get out more [Interviewee L]*

Other reported changes were efforts to improve general wellbeing through making more time for sleep, taking more exercise and attending more closely to diet.

While these interviewees were clear on the need for, and likely benefits of, change, many also identified barriers and challenges. We do not know how successfully these were circumvented, and whether all the intended changes were ultimately made. Although challenges were seldom framed as insurmountable, the need for support was emphasized. Challenges to initiating and maintaining the recommended behaviours were diverse, relating to: other symptoms and/or conditions; personal responsibilities and resources; individual psychology; and the clarity and immediacy of the "return" on the changes. One interviewee warned:

> *If it doesn't work in the first week, and make an, an instant difference, it's difficult to, to just really stick with it. [Interviewee E]*

We noted that nobody reported any change in fatigue or its impacts by the time of the second interview. By contrast, interviewees often stressed that fatigue continued to impact negatively on their quality of life. Professionals attending the focus group said that this was to be expected, and that patients should be warned that in the short-term their sense of fatigue might even increase:

> *One of the real blocks to people gradually doing more is the belief that hurt equals harm… They think, "Oh, my symptoms have got worse, I should stop"… You (need to) warn people that they'll get worse (before they get better). [FGP U]*

3.3 | Contribution of the research experience

Research participants (interviewees and focus groups members alike) saw the research project as providing a distinctive context for exposure to the booklet. In several instances, interviewees cited this as significant. The data more generally suggest three ways that the research process may have contributed to the changes in thought and behaviour reported. Firstly, it established the relevance of the booklet (with participant information documents explicitly linking fatigue and arthritis, and recruitment conversations reinforcing this). This was of obvious importance where interviewees had not previously connected fatigue with their condition. It was also helpful to those who had not named their experience "fatigue" or whose diagnosis did not feature in the booklet title:

> *What you're always looking for is something specifically about you… (And) it doesn't say ankylosing spondylitis anywhere on there. [Interviewee E]*

Secondly, the line of questioning adopted in the "pre-booklet" interview prompted patients to reflect on their current approaches to fatigue management. Interviewees were asked to describe, in some detail, their own strategies for managing fatigue and how effective these had been. The use of "How", "What if" and "Why (not)" questions introduced gentle challenge. For several interviewees, this led to an acknowledgement that their current approach to managing fatigue was sub-optimal, a logically necessary precursor to contemplating change. The third significant feature was commitment to follow-up, in the form of the "post-booklet" interview. Being held to account was cited as important by several interviewees:

> *If you hadn't been coming back would, would I have actually sat down and read the book from cover to cover, and actually, you know, give it the concentration I did? I probably wouldn't, I prob-, I probably wouldn't, to be honest. [Interviewee H]*

Focus group members also saw accountability as key:

> *With any information-giving, it needs to be reviewed. [FGP Z]*

Focus group members considered the potential for these research features to be reproduced in routine practice. The group agreed on the importance of rheumatology professionals drawing attention to the association between fatigue and rheumatic disease, and the potential to manage it using non-pharmacological strategies. They suggested that the *Fatigue and arthritis* booklet could be introduced effectively in the context of such a conversation. They viewed it as both desirable and feasible to introduce an element of accountability (and advocated adding a template to the booklet for recording intentions or goals in support of this). They suggested that professionals could, and should, commit to discussing the booklet and patients' goals at future appointments; patients might be encouraged to identify a friend or family member who could hold them to account in the interim.

4 | DISCUSSION

Recent years have seen information provision play an increasingly prominent role in health policy (Department of Health, 2012; Department of Health and Human Services (US), Office of Disease Prevention

and Health Promotion, 2016; Washington & Lipstein, 2011). It has been conceptualized both as an intervention in itself and a central plank in shared decision-making initiatives (Elwyn et al., 2010) and self-management programmes (The Health Foundation, 2015). High-quality information has been described as that which is relevant, evidence-based, developed with users and embedded within care (Patient Information Forum, 2013). Increasingly, the case for investment in health information draws on "discourses" (Dixon-Woods, 2001) of both patient education *and* patient empowerment. It has been argued to improve quality of care, service use and costs, patient outcomes and patient satisfaction (Department of Health and Human Services (US), Office of Disease Prevention and Health Promotion, 2016; Patient Information Forum, 2013).

Written information has been characterized as low-cost, flexible and an effective aid to understanding and recall (Ellis, Hopkin, Leitch, & Crofton, 1979; Harris, Smith, & Veale, 2005; Patient Information Forum, 2013). However, some authors have warned that care should be taken not to overstate its effects and cautioned that different patient groups may not benefit equally (Blickem et al., 2011; Thompson, 2011). Ongoing disparities in access to written information in electronic form remain a concern to policymakers (Department of Health and Human Services (US), Office of Disease Prevention and Health Promotion, 2016). Furthermore, it has been questioned whether information alone can be relied upon to bring about behaviour change, and argued that theoretically grounded behavioural programmes have better outcomes (John et al., 2011). A recent publication by The Health Foundation (2015) reached the conclusion that information may increase knowledge, but that to influence behaviour other forms of support may be needed.

At first sight, our own research, which finds written information to have an impact both on thoughts and behaviours, appears at odds with this wider evidence base. However, taking into account the context in which the booklet was distributed and how, in consequence, people engaged with it, our findings become easier to reconcile. We explain this in more detail below. Then, having specified the conditions under which the booklet brought about change, we conclude by making some suggestions as to how its impact might be reproduced.

While it is clear that people need to encounter the booklet, they may either find it or be given it. Our data suggest that there are advantages to the latter, and that when people are given a booklet by a professional (in the case of our project, a researcher), they engage with it more actively. This resonates with claims made elsewhere (Thompson, 2011; Patient Information Forum, 2013) regarding the value of what the latter organization terms "infomediaries". We have already noted how some patients with conditions other than (rheumatoid) arthritis expressed uncertainty, initially, as to whether the booklet was intended for them. In addition to confirming its relevance, professionals can frame engagement with the booklet in a number of important ways. In the present study, this included inviting patients, in the initial interview, to reflect on their prior experiences and management practices. In many instances, this led to recognition that their management practices were sub-optimal and might be modified. It seems likely that this may be a pre-condition for behaviour change. The Health Foundation (2015) have reported that the impact of written materials (and, indeed, of other forms of information and support) on self management, is maximized when backed up by professionals using techniques such as motivational interviewing (Elwyn et al., 2014; Treasure, 2004) to help patients to develop goals and solve problems in the course of consultations. Although the research interviews were not intended to take the form of motivational interviews, a key feature of that type of counselling – the expression of empathy through reflective listening (Treasure, 2004) – was a characteristic. In particular, the second interview provided a forum for people to reflect on their practices and how the advice contained in the booklet fitted with, had or might affect these. The serial nature of the interviews was, perhaps, the most significant feature of the research, with all participants expecting to be asked to give an account of their reaction to, and use of, the booklet. The role of follow-up in supporting behaviour change is acknowledged in the literature (Sohl, Birdee, & Elam, in press). Active and sustained follow-up of patients' self-management goals (in addition to their clinical status) is also a key component of the "productive interactions" (Cramm & Nieboer, 2014) associated with Wagner's chronic care model (Wagner, 1998).

These features of the research are all potentially replicable in routine practice, and at relatively little cost. This is an important pragmatic consideration in the UK, where growth in demand for health services has not been matched by increases in resources (Roberts, Marshall, & Charlesworth, 2012). Health professionals, however, may themselves need support to make the most effective use of information materials such as the fatigue booklet (Department of Health and Human Services (US), Office of Disease Prevention and Health Promotion, 2016). Almost 20 years ago, after a wider review of educational materials for patients with arthritis, Bishop et al. (1997) stressed the importance of educating professionals in the use of patient literature, and guidelines on how to make the most effective use of leaflets were produced. Our findings suggest that there might be value in updating those guidelines and actively encouraging health professionals to use literature such as the *Fatigue and arthritis* booklet to support and enhance their patients' care.

4.1 | Limitations of the study

We do not deny that our research has its limitations – most obviously, study duration and sample (size and character). Data regarding the impact of the booklet on patients' thoughts and/or behaviours do not suggest any difference by diagnosis. More nuanced differences in patient characteristics (e.g. education) and circumstances (e.g. life demands) may be significant, but our sample does not allow us to reach firm conclusions on this. There remains scope to characterize further the exact population to whom the benefits of the booklet – with and without additional support – might extend. Another important question is the extent to which reported benefits are sustained (and what type and level of intervention might promote this). However, notwithstanding these limitations, we believe that our research indicates that the potential of written information on fatigue and rheumatic disease is yet to be fully realized, and offers some useful pointers as to how such resources might be used to greater effect.

5 | CONCLUSION

Fatigue is a common symptom of autoimmune rheumatic diseases, with a significant impact on health-related quality of life. Patients struggle to understand this symptom and get little support to manage its effects. Written information, in the form of a booklet, can change how patients think about fatigue. This is valuable, alleviating a range of concerns and equipping them to improve their management behaviours. Dissemination of written information by professionals, guided reflection with sensitive challenge, and a clear commitment to follow-up encourage patients to convert changes in thought to changes in behaviour. For maximum effect, written information needs to be embedded within the conversations and practices of routine outpatient care. Used in such a way, it offers a low-cost tool for addressing as yet unmet patient needs.

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Clinician–researchers and custodians of scarce resources: a qualitative study of health professionals' views on barriers to the involvement of teenagers and young adults in cancer trials

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Abstract

Background: Equipoise and role conflict have been previously identified as important factors in professionals' engagement with trials, inducing behaviours which can impact on recruitment. We explored these phenomena as potential explanations for the low levels of involvement of teenagers and young adults (TYA) with cancer in clinical trials in oncology.

Methods: We report findings from interviews with 30 purposively sampled direct care professionals involved in delivering cancer care and/or facilitating clinical trials in Scotland. We undertook qualitative descriptive analysis, focussed on identifying key issues and themes.

Results: Interviewees largely identified as clinician–researchers and portrayed oncology as a specialty in which research was integral to care. They saw their primary responsibility as ensuring patients received the best treatment, but asserted that, in general, trials provided a vehicle for optimal care. Role conflict in its traditional form was rarely evident; however, other tensions were manifest. Professionals found the significant time costs of delivering trials difficult to reconcile with the increasing pressures on clinical services. They felt a responsibility to make prudent choices about the trials with which to engage. Guided by utilitarian principles, these choices were oriented towards benefiting the largest number of patients. This favoured trials in high volume diseases; as TYA tend to have rarer forms of cancer, professionals' support for—and TYA's access to—relevant trials was, by default, more limited.

Conclusions: Neither lack of individual equipoise nor experiences of traditional forms of role conflict accounted for the low levels of involvement of TYA with cancer in clinical trials. However, prominent tensions around the management of scarce resources provided an alternative explanation for TYA's limited access to cancer trials. The prevailing approach to decision making about whether and which trials to support was recognised as contributing to inequalities in access and care. Professionals' choices, however, were made in the context of scarcity, and structured by incentives and sanctions understood by them as signalling governmental priorities. A franker discussion of the extent and distribution of the costs and benefits of trials work is needed, for change to be achieved.

Keywords: Trials, Equipoise, Role conflict, Teenagers & young adults, Cancer, Qualitative research, Interviews

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Background

Clinical trials have played a definitive role in medical progress, particularly in the field of cancer care [1]. Important patient outcomes, including survival, have improved significantly in that specialty since the expansion of trial activity in the mid-twentieth century [2]. However, not all cancer patients have had the same level of involvement in trials, and patient populations with lower levels of trial involvement have seen more modest improvements in outcomes. Teenagers and young adults (TYA) with cancer are one such group; levels of trial participation and improvements in outcomes for this group contrast particularly dramatically with those achieved in paediatric oncology, where trial participation levels have been high [3–5].

Whilst clinical trials are powerful tools for advancing knowledge and patient care, their successful delivery is difficult, with recruitment challenges affecting timely completion of a significant proportion of studies [6, 7]. A growing body of research has sought to understand the barriers and facilitators to recruiting into clinical trials, initially by exploring patients' views and experiences [8–13]. More recently, researchers have investigated the perspectives of health professionals charged with recruiting, consenting, and retaining patient participants [14–17]. Two distinct but related explanatory concepts have been suggested as affecting professional recruitment practices and trial outcomes: the existence (or not) of equipoise and experiences of role conflict.

Clinical equipoise [18] is considered a fundamental ethical requirement of any trial involving random allocation. In essence, being 'in equipoise' means being in a state of uncertainty about the relative scientific or clinical merits of the different arms of a clinical trial. When there is uncertainty about which treatment is preferable, then conducting a trial is justified. Distinctions have, however, been drawn between clinical and individual equipoise (uncertainty), and these may not be fully aligned [15, 19]. So, while health professionals may agree that clinical equipoise exists, i.e. recognise that a lack of consensus exists amongst medical experts, equipoise at the individual level is often less stable. Individual healthcare professionals may believe that one form of treatment is generally superior, or that specific (groups of) patients would benefit more from one treatment than another. Disturbances in individual equipoise may lead professionals to undertake selective or biased recruitment, such as not broaching trial participation with certain groups or individuals, or introducing a trial in a way that prompts patients to decline involvement [14, 19–21].

Underpinning these behaviours is a concern that participation would not be in patients' best interests. The care obligations at the heart of the clinician-patient relationship are deeply felt, and whilst clinical equipoise may provide ethical justification for allocation of patients to treatment according to a trial protocol, individual uncertainty will almost inevitably leave clinicians feeling conflicted. Broader tensions between goals and responsibilities relating on the one hand to research or scientific endeavours, most particularly clinical trials, and on the other to the care of (individual) patients, have been conceptualised as manifestations of role conflict [22–24]. Role conflict arises because clinical trials are driven by hypotheses, which are operationalised in protocols that determine the care a patient receives (in lieu of assessments of patients' individual circumstances, preferences or needs). This can lead professionals to question whether the interests of their patients might not be better served by more tailored care, albeit that the provision of such care may not be consistent with the priorities of the trial. This sort of conflict has been identified as generating significant discomfort for health professionals in a range of positions and specialisms. As with lack of equipoise, where responsibilities to individual patients are deemed to conflict with and override those due to research endeavours, behaviours which may impact negatively upon recruitment can be triggered [20, 25, 26].

As part of a wider study of barriers (and facilitators) to TYA's involvement in cancer trials, we interviewed health professionals involved in delivering cancer care and/or facilitating clinical trials. In keeping with the literature, we anticipated that a lack of equipoise (individual uncertainty) and/or role conflict might offer explanatory vehicles for the low levels of involvement of TYA in cancer trials. Our findings did not support this. In particular, they (our findings) did not show role conflict, as currently understood and employed in the literature, to account for limited trial participation by TYA. However, as we explain below, they revealed other new and highly significant tensions, with ultimately similar consequences for TYA's involvement in trials.

Methods

We outline our methods—and findings—broadly in accordance with the consolidated criteria for reporting qualitative studies (COREQ) [27]. The use of such checklists is advocated in and by many clinical and scientific journals [28], although it has been problematized by some prominent researchers [29].

Design and theoretical framework

We undertook a qualitative study, involving one-off semi-structured interviews with direct care and other professionals, patients and family caregivers (data from non-care professionals, patient and caregiver interviews will be reported separately in due course). The study was

characterised by an emergent, inductive design, purposive sampling and an iterative approach to data collection and analysis [30]. This iterative approach enabled us to revise our sampling strategy and interview topics, and to capture and unpack emerging issues (including some unanticipated when the study began). Our work was lightly informed by Normalization Process Theory (NPT), a middle-range theory offering an explanatory framework for 'how complex practices… are made workable… in context-dependent ways' (p.536) [31]. Amongst other things, NPT sensitised us to the potential for differing institutional contexts to shape and produce variation in interviewees' experiences and perspectives, and the need to allow for this possibility in our sampling and interview questions.

Setting

We conducted our study in Scotland. Health is a devolved matter, with legislation relevant to the National Health Service (NHS) in Scotland made by the Scottish Parliament. NHS Scotland is legally and financially independent from NHS England and has a distinct organisational framework. Fourteen regional 'Health Boards' plan, commission and deliver services, in line with the Scottish Government's priorities. Major and specialist facilities are concentrated in the four largest health boards, although more than 20 hospitals in Scotland provide cancer care. Research infrastructure is provided by NHS Research Scotland (NRS), which takes its strategic direction from the Chief Scientist Office (CSO), an arm of the Scottish Government (and the funder of this research), and the four largest health boards.

Participants and recruitment

Our approach to sampling was purposive and focussed on construction of a sample reflecting the diversity of relevant experiences and perspectives. Hence, we set out to recruit medical and nursing staff from different subspecialties, types of service, hospitals and health boards (for example, small, large, rural, urban, with and without specialist TYA services/facilities). In light of the issues emerging in the first phase of the interviews, which related substantially to what might be termed structural factors (obstacles to the timely approval and set-up of relevant trials), we later sought to recruit professionals whose roles gave them particular insight into those matters. Initially, potential research participants were identified by clinical members of the research team. As sampling progressed, help to identify professionals with particular characteristics and/or experiences was sought from advisory group members and other clinical colleagues. We also invited interviewees to suggest colleagues with whom we might usefully speak. Interviewees were not asked to introduce us to their colleagues and were not informed whether their suggestions were followed up.

Approaches to potential interviewees were made in writing, either by the Chief Investigator (AJ) or the project researcher (RH), and were followed up by the latter. All potential interviewees were given detailed participant information sheets and invited to opt-in by replying directly to the project researcher. Approximately half of those approached agreed to take part. Where reasons were given for declining participation, these typically related to pressures of work and/or the small number of TYA patients seen. We discontinued recruitment once the team was in agreement that sampling ambitions had been satisfied and data saturation had been reached (i.e. the data set included an appropriate range of participants and perspectives, and additional interviews were no longer generating new issues or themes).

Data collection

Data was collected between December 2017 and August 2018. Interviewees were given the option of either a face-to-face or telephone interview, with the former (16/30) typically taking place in the interviewee's own office or a private meeting room. These were conducted by the project researcher (RH), a social scientist with prior experience of undertaking qualitative health research in haemato-oncology and other clinical areas.

Interviews were semi-structured, loosely following a topic guide outlined in Table 1. This was informed by a review of the literature, the expertise of the research team, and input from advisory group members. In line with study's inductive and iterative approach, the topic guide was revised over the course of the study in light of emerging findings. Questions were modified in situ to take account of interviewees' varying roles, experiences and perspectives. As such, the *guide* should be considered precisely that, and not as a script. Interviews typically lasted 45–60 min. All were audio-recorded and then transcribed verbatim. After anonymization had been fully confirmed, transcripts were imported into the specialist data management software, NVivo (Version) 11 (QSR International Pty Ltd., Doncaster, Victoria, Australia).

Data analysis

We undertook qualitative descriptive analysis, with a focus on producing rich, 'minimally theorized' descriptions of views and experiences [30, 32]. After initial familiarisation with the transcripts, which involved close reading and line-by-line coding, two members of the research team (RH and JL) undertook more focussed coding and 'constant comparison' [33]. Having undertaken independent analyses of the data, these researchers met to discuss and reflect upon their interpretations. The two analyses were very similar; hence,

Table 1 Interview topic guide

About the interviewee:
Role/responsibilities, including clinical and other interests
Contact with TYA patients
Experiences of providing care to TYA
If/how providing care to older adults (or children) is different
Involvement with trials:
Understanding of the term
Experience of trials work
Reasons for doing trials
Views on costs and benefits of trials (to current and future patients, professionals, and organisations)
Decision making about whether to open any specific trial locally
Processes for opening a trial locally
Barriers and facilitators to timely opening of trials
Scope for improving processes
Recruiting TYA (and other) patients to trials open locally
Experience of recruiting TYA (and other) patients
Processes for identifying eligible patients
Deciding whether or not to approach individual patients
Who/what is involved in recruitment
Issues, concerns, and/or challenges specific to TYA (in general and/or sub groups)
Referring TYA (and other) patients to trials open elsewhere
Experience of referring TYA (and other) patients
Processes for identifying eligible patients
Deciding whether or not to approach individual patients
Who/what is involved in referral
Issues, concerns and/or challenges associated with referral to trials open elsewhere
Issues, concerns and/or challenges specific to referring TYA to trials open elsewhere
Increasing TYA participation in trials
Explanations for variation (in levels of participation) between services and sub specialisms
How to promote trial participation by this age group
Other issues or comments
Changes in context and/or trials
Emergent issues

consensus on the coding frame for subsequent work was easily reached. The final stage of the analysis involved a further round of coding and constant comparison, with the project researcher (RH) systematically comparing, sorting and associating data and codes, until patterns, subtle variations and exceptions became clear. Summaries, diagrams and analytical reports enabled the analysis and gave other members of the team access to the data and the analytical logic.

Results

We interviewed 30 direct care professionals whose role(s) involved delivering cancer care and/or facilitating clinical trials in Scotland. Participant characteristics are detailed further in Table 2.

In the sections that follow, we report how (1) these professionals largely identified as clinician–researchers and offered little evidence of experiencing role conflict in its traditional form, but (2) other powerful tensions had emerged around the use of scarce resources. Quotes are attributed to participants who are distinguished using unique alphanumeric codes, the initial letters of which provide information on their role: DC-C/N/O (Direct Care-Consultant/Nursing/Other). No relevant differences in perspective by role/staff group emerged.

Identifying as clinician–researchers

Interviewees with direct care roles strongly identified as clinician–researchers, with accounts emphasizing the close integration between research and care in contemporary oncology. They portrayed involvement in research, particularly clinical trials, as an integral part of their work; something which was the norm, rather than the exception, for professionals within their specialism:

> 'it's part of core work… you can't do oncology without doing research, it's just… part of our job… just about every oncologist is involved in clinical trials… and if you're not, you've got the question, why not?' (DC-C-11).

> 'in oncology, (research) is integral to clinical care… that's what makes it such an interesting speciality, everyone's involved… (and) it's very integrated into clinical practice.' (DC-C-12).

Clinicians described being introduced to clinical trials work early in their medical career and talked of taking on more responsibilities for trials work as they gained experience and status in oncology. Current oncology trainees, they said, were being prepared to take on similar responsibilities in the future. For example, as part of the medical oncology curriculum, trainees were all expected to undertake 'GCP' (Introduction to Good Clinical Practice) training, and to be involved in recruiting patients to trials, as well as providing follow-up care.

Some nursing interviewees were in research/trials nurse roles at the time of interview; others were not, but had held such posts in the past. Several recalled involvement, as staff nurses, in the care of cancer patients treated through clinical trials. This, they said, had given them too a strong sense of the importance of trials work and its contribution to improving cancer care:

> 'you can see the development of drugs over the course of the years… you can actually see that with, you know, with your own eyes… that's motivational, you

Table 2 Participant characteristics

Interviewees with direct care responsibilities		30
Type of healthcare professional	Consultant	23
	Nursing	4
	Other	3
Consultants' specialisms	Clinical oncology	7
	Medical oncology	6
	Haematology	6
	Paediatric medicine	3
	Surgery	1
Consultants' site/sub specialisms*	Haematological malignancies	5
	Gastro intestinal cancers	4
	CNS/Neurological	3
	Genito urinary cancers	3
	Breast cancer	2
	Gynaecological cancers	2
	Head, neck & thyroid cancers	2
	Sarcomas	2
	Paediatric oncology	2
	Lung cancer	1
	Melanoma	1
	MCUP	1
If also had research development or support responsibilities	No	25
	Yes	5
Type of responsibilities	Senior role in a CTU or CRF	3
	Specialist trial support**	2
Board to which primarily affiliated	NHS Lothian	11
	NHS Greater Glasgow & Clyde	8
	NHS Highland	4
	NHS Tayside	3
	NHS Grampian	2
	NHS Fife	1
	Other primary affiliation***	1

* Several consultants worked in more than one sub specialism; therefore, these numbers do not sum to 23
** With regard to pharmaceutical or radiographic components
*** Interviewee primarily affiliated to an institution outside Scotland

know, to being positive about trials, and taking part in them.' (DC-N-10).

Both clinicians' and nurses' accounts gave a strong impression of a trial culture, underpinned by a commitment to evidence-based medicine and with a focus on improvement, patient safety and the provision of the highest quality of care:

'if we don't do clinical trials, we will make decisions… not based on the best evidence… we have to do clinical trials to actually improve outcomes, but also to protect patients from drugs or interventions that might actually not be as effective as what's already available.' (DC-C-26).

Significantly, clinical trials were viewed not only as delivering benefits for future patients, but also as profiting current patients. Interviewees largely shared the opinion that research-active organisations achieved better results for patients (generally) and that patients who took part in trials had superior outcomes to those who did not. Interviewees acknowledged some uncertainty as to why patients in centres that delivered trials did better. They

offered explanations concerning, inter alia, the culture or outlook developed by involvement in trials, and the value of external scrutiny of practice:

> 'Trials make people think… (be) adaptable and willing to change for the better… that's a culture that… taking part in clinical trials develops, or encourages.' (DC-N-10).

> 'we also know that… being part of research is quite good from a quality control point of view… your patient care… is being compared by the trials monitoring committee, it's almost like… intellectual peer review… of practice.' (DC-C-16).

A significant proportion of interviewees referred to 'evidence' suggesting that patients enrolled in trials had better outcomes. Again, interviewees acknowledged it was not yet entirely clear why trial-enrolled patients did better. They stressed that new treatments or new ways of administering treatment did not always outperform standard care and cited 'research' showing that control arm patients also tended to have better outcomes than non-enrolled peers:

> 'there's… evidence… that patients who participate in trials do better, irrespective of whether they've got the trial treatment.' (DC-C-13).

Interviewees suggested that the explanation was probably 'multi-factorial' (DC-C-11), identifying a series of factors or mechanisms for what is sometimes referred to in the literature as 'the trial effect' [34]. These related largely to care practices—careful observation of a treatment protocol, scrupulous monitoring, and diligent recording:

> 'there's quite a lot of evidence to say that patients in clinical trials get better clinical care, just 'cause they are seen more frequently, and so you're much more likely to pick up on symptoms, and problems, at an earlier stage. So for individuals, it's always important to stress that the benefit's not necessarily from the drug, or the technique, because we don't know yet if that's going to make any difference, but we know that generally speaking, because of the more intensive care provided for people in trials for collecting all the data, that translates into sort of better clinical care.' (DC-C-5).

Whatever the explanation, treatment through a trial was, in principle, viewed as giving patients access to the best possible care. As such, our interviewees gave little impression of being conflicted in the performance of their clinical care and research responsibilities; in contrast, they saw trials as a vehicle through which they could offer their patients optimal clinical care.

Accounting further for the apparent lack of conflict between care and research, interviewees reflected on the characteristics of the trials with which they were involved, and the distinctive nature of those of most relevance to TYA patients. Although some direct care interviewees reported involvement in early phase trials, most interviewees and accounts focussed on 'Phase 3' (large-scale, randomised) trials of first-line care. These are the sorts of trials involving the largest number of patients and, archetypally, compare two quite distinct drugs, procedures or technologies, one of which is new. The trials described by our interviewees appeared notable for their complexity, comparing differences in protocols which could involve multiple treatment modalities. Interviewees acknowledged that, *in principle,* such a trial might still expose patients to a new agent that was inferior to current care. However, several interviewees took pains to note that 'new' did not mean entirely unknown, one commenting as follows:

> 'in (this sort of) trial you feel you've come far down the road enough, for, you know, the "new" drug not to be a worse drug. It (may) not be better, but there are reasonable data to say… it shouldn't be worse.' (DC-C-7).

They stressed that patient safety was the prime consideration in the design of trials and that all were very carefully scrutinised before initiation. Though laborious to navigate, regulatory and review processes functioned both to minimise harm and to reassure clinicians that they were not exposing their patients to inappropriate risks:

> 'Our process for taking on trials, and the way that our consultants work… we're quite confident that we're not offering things to people that wouldn't be good for them.' (DC-N-10).

Moreover, interviewees described trials in the disease areas of most relevance to TYA patients (for example, sarcomas, brain tumours, and acute leukaemia) as commonly exploring different ways of using familiar treatments, sometimes using 'different treatment approaches using the same drug' (DC-C-9), rather than investigating the effects of new ones:

> 'Some of the studies… are actually looking at what other medicines we can take out, or lower the dose of, to cut down on side effects, so… trials are not necessarily about *new* treatment.' (DC-C-17).

> 'Trials (can) offer an opportunity to access medicines that we otherwise wouldn't be able to… (But) a lot of our trials… are not like that though… a lot of our trials involve making minor modifications to… standard treatments.' (DC-O-24).

As the quotes above suggest, interviewees portrayed these trials, which seldom had commercial funding, as often looking at quite subtle modifications to care, for example, making adjustments to treatment combinations, order, dosage and/or frequencies. Interviewees talked of attending to 'the finer details of the treatment… trying to clarify aspects of that treatment' (DC-C-3). One explained how they proceeded:

> 'building on what our, our gold standard, or our best available therapy is, and we're trying to tweak the treatment slightly to improve outcomes, or add in a new treatment, or change the doses, or change the scheduling of it… (to) further improve the treatment… to improve the outcomes.' (DC-C-17).

Priorities varied between disease areas / tumour types, but outcomes of interest included both survival and (short and longer-term) toxicities. The latter was viewed as of particular significance to TYA patients:

> 'in both haematology and breast cancer… we've got such good outcomes really, that it's all about trying now to look at, how little do we need to do to get the same outcomes, to keep the quality of life? So there's quite a lot of work about how much can we pull back… and still achieve the same results… The younger population, those people are hopefully going to be cured, and then live till they're 80, so you don't want them to live with the side effects of the chemotherapy or radiotherapy' (DC-C-15).

Such trials were framed as exposing TYA patients to worthwhile benefits and quite limited risks:

> 'I'm fairly enthusiastic about putting my acute leukaemia patients in trials… because… they get a fairly good deal in these trials and the risk-benefits… are pretty good.' (DC-C-7).

Furthermore, the nature of these trials meant that the superiority of any particular arm was exceedingly difficult to pre-judge:

> 'the basis for the trial is that we don't know which one's going to be better, or better tolerated… that's the nature of a Phase 3 trial, we can't run it unless we think there's genuine uncertainly about which one is best. (And in these trials) that difference is much less apparent to anybody, even (those) running the trial. It's less clear what, whether you would have been better off, if you had the other bit.' (DC-C-3).

New tensions emerging

Whilst interviewees expressed a deep commitment to research and saw it as an integral part of their responsibilities, most were employed primarily to deliver clinical services:

> 'most people in, in oncology are NHS doctors… who believe in research, who do research, but whose first commitment is to the NHS service.' (DC-C-11).

> 'although they are interested in research, and involved, and want to... be involved, they have a clinical workload… real patients, and real people, that have to be dealt with.' (DC-O-30).

Many talked of growing pressures on core services: clinician and nursing time, pharmacy, radiology/radiotherapy services and laboratory staff and facilities. Certain fields (solid tumour oncology), services (radiology), and health boards (rural) were suggested as facing particular or persistent challenges:

> 'in adult solid tumour oncology, we've had a severe shortage of staff… across the UK. So workload has been a huge issue…' (DC-C-25).

Interviewees from one health board reported longstanding difficulties with clinical staffing, with vacant consultant posts leaving them heavily reliant upon locums. Meanwhile, colleagues in another health board described their pharmacy and radiology services as struggling to deliver standard care. They relied on their larger neighbours for some work related to routine clinical services:

> 'we… use (City) labs for a lot of things anyway… that's standard… if there's extra tests… that can be an issue.' (DC-N-22).

These pressures made the costs associated with undertaking trials, in particular staff time, increasingly hard to absorb. Although trials were framed by some interviewees as a potentially useful source of income, they were more frequently construed as drawing critical resources away from routine care. This may in part be reflective of the source and levels of funding for the TYA-relevant trials around which conversations revolved. All current and recent trials of clear relevance to TYA

with cancer which featured in interviews were non-commercial, i.e. funded by charities or through (European) grant programmes.

Notwithstanding their commitment to participating in such research, many interviewees talked at length of the significant monetary and other (e.g. in-kind) costs associated with establishing, opening and maintaining these trials: 'trials are demanding in time, they're demanding in resource…' (DC-C-16). They drew attention to the substantial time costs associated with both opening and delivering a trial. These costs included trial administration, recruitment, delivery of complex trial interventions, data collection and reporting.

The demands associated with trial administration received particular attention and were framed both as significant and expanding. An experienced clinician remarked how, over their professional lifetime, they had 'ballooned, absolutely ballooned' (DC-C-25). Comments were also made that whilst non-commercial trials used to be more straightforward to administer, they were now expected to meet the same regulatory and reporting requirements as commercial trials—but without equivalent resources and infrastructure. Opening a trial was portrayed as extremely laborious and, critically, due to the requirement to maintain and update documentation to reflect (increasingly frequent) amendments, the ongoing demands on staff time were also considerable:

> 'Seven or eight years ago, you would maybe see one or two protocol amendments in the lifetime of a study. Now you're seeing two or three a year, it's out of control, there's more protocol amendments than patients on half our studies. And staying on top of that is extremely difficult.' (DC-O-9).

Interviewees emphasised that, in large part, these administrative demands bore no relationship to the scale of recruitment to the trial. Due to broadly equivalent costs of activation, set-up and maintenance, trials where recruitment might be expected to be limited (often the case with those in disease areas relevant to TYA) could prove as much a drain on resources as those with much higher levels of recruitment:

> 'if we do a trial where we're going to recruit 10 patients, then obviously the amount of work that I have to do as a PI… and some of the nurses have to do – in terms of bureaucracy – is actually pretty much the same as if we were to do a trial where we might only recruit one.' (DC-C-25).

Hence, in this context of high demand and scarce resources, accounts suggest that trials were in competition not only with clinical services, but also with each other. Interviewees drew attention to the 'finite' nature of resources, the 'opportunity cost' of trials work, and the consequential requirement to make prudent choices about *which* trials to engage with and fight for support for:

> 'as a department, we've got limited research resource, so we… focus on trials… (with) a chance of benefiting our patients. Focus our nursing and data management resource into that.' (DC-C-11).

Pressed to explain further how decisions were made whether to engage with (i.e. open) a particular trial or not, interviewees talked about assessing if a trial was 'worthwhile'. This appeared to mean a range of different things, prominent amongst which was recruitment potential:

> 'it's about numbers, how many people do you need to get into a trial in order to make the trial… worth the resource…?' (DC-C-3).

Professionals appeared to evaluate trial opportunities according to the principles of utilitarianism, which, at its simplest, is an economic philosophy oriented towards achieving the greatest good for the greatest number of people. Interviewees acknowledged that the prevailing value system privileged high volume diseases (for example, breast and prostate cancer). As one interviewee put it, 'the big tumour types… dominate' (DC-C-11). Accounts suggest that the system of rewards and penalties for trials work established by the Scottish Government (CSO) had encouraged and embedded such an approach. Its use of recruitment figures as the central metric incentivised trials in those centres, disease types, subtypes and phases where there was greatest potential to recruit:

> 'Not to beat around the bush, we get very little money, until we recruit patients. You know, most of the incentivisation… bean-counting, is related to the numbers of recruits, not to the numbers of… studies. And so, you know, those are resources which, we're putting in, where we may get nothing back, and would frankly be better directed to something where we're gonna get something back.' (DC-C-28).

Some interviewees questioned the rationale of the contemporary rewards system, musing that:

> 'number of recruits to studies… that's a slightly lazy way of trying to appreciate the quality and quantity of patient benefits… if you run a study on relatively few patients, but actually that's a novel, good intervention,

> that would be a great benefit… the health impact may be… more significant that a study that runs on 100 patients which provides relatively minor, additional health benefits… Yet the 100-patient (study) would win, in the eyes of the Government.' (DC-C-27).

Ultimately, and notwithstanding ambitions to have trials available for as many different patient groups as possible, there was widespread recognition that, in reality, only a minority of patients aged 16+ had access to care through a trial. One interviewee (DC-C-20) put it very bluntly: 'for the majority of patients, there isn't a trial.' Significantly, however, the prevailing logic and rewards metric led to patterned inequalities in access to trials, powerfully dis-incentivising the opening of trials relating to rarer cancers such as those with which TYA typically presented:

> '(be)cause the numbers are small… staff are kind of dis-incentivised… A lot of people just say, I just can't be bothered with that if I'm only gonna recruit, you know, one patient in six months or something, why the hell do I wanna do that? I've got other things to do.' (DC-C-25).

Some interviewees highlighted a wider lack of trials in rare tumours, framing pharmaceutical companies as having limited interest in developing and trialling new treatments for such diseases (as the returns were likely to be modest and only distantly realised). Others questioned the extent to which smaller centres, outside London, were given an opportunity to participate where such trials were established. Many, however, explicitly acknowledged the role—and consequences—of local decision-making:

> 'There is a national trial for rhabdos, but we haven't opened it in, the adult site, because it's a very complicated trial, and we would probably only get one patient every two or three years… 'cause it only goes up to (age) 21, I think' (DC-C-3).

> 'there are some trials for very rare cancers… quite a few that are relevant to this group (TYA), where we have had to just make the decision, we don't have enough resources to open this trial that we may recruit one, or zero patients (to) over the lifetime of the trial… that's a big problem… for this group of patients.' (DC-C-18).

It seems TYA patients might not be deprived of trial opportunities by their age per se, but by their tendency to present with tumours uncommon in the wider adult population:

> 'we've got about 160 trials open… (but) there's probably about half a dozen… which would include diseases which were relevant (to TYA).' (DC-C-28).

Discussion

A key finding of our research is that interviewees identified strongly as clinician–researchers and portrayed oncology as a specialty in which research was integral to care. Examples of role conflict in its traditional form [22–24] were largely absent from our data. However, there was evidence that new and significant tensions had emerged, which offered a powerful alternative explanation for the low levels of involvement of TYA with cancer in clinical trials in oncology. Specifically, our data suggest that, inasmuch as research was felt to conflict with care, it was in its potential to consume increasingly scarce and precious clinical resources. Our data reveal an acute appreciation of resource scarcity and a sense of obligation amongst professionals to make deeply pragmatic choices about those trials in which to invest their time. Guided by utilitarian principles, these choices were oriented towards benefiting the largest number of patients. This favoured trials in high volume diseases; as TYA tend to have rarer forms of cancer, their access to trials was, by default, limited. Interviewees recognised that the choices they made as professionals about which trials to support had very concrete repercussions for equality of access—and arguably care. TYA and other patients presenting with rare cancers (and indeed other rare diseases) were acknowledged as being systematically disadvantaged.

These findings do not fit entirely easily with the existing literature. Traditionally, the roles of clinician and researcher have been understood to be fundamentally distinct and inherently conflicting. Attention has been and continues to be drawn to differences in the goals, practices, responsibilities, obligations, risks and ethical frameworks for research and care [20, 22–24, 26]. The form and consequences of these role conflicts (for clinicians, patients and, to a lesser extent, researchers) have been relatively well documented, including in studies focussed on the conduct of clinical trials [14, 16, 35]. Role conflict has been found to have implications for both recruitment and trial delivery; for example, previous work has shown that clinicians will not follow a trial protocol if they are concerned that it will deprive their patients of the best clinical care [36].

That said, recent years have seen the publication of a growing body of work which challenges the conceptualisation of care and research as unerringly distinct and conflicting. That work suggests a more permeable boundary between research and care, at least in some technical specialities [37–39]. Keating and Cambrosio

[1] and Cambrosio et al. [40] have argued that, in the field of oncology, the practices of standard care and participation in trials have become closely intertwined. Our own work very much supports that claim. Whilst interviewees were clearly cognisant of the clinical essence of their identity [20] and saw their primary responsibility as ensuring patients received the best clinical care, they also viewed trials, in general, as a vehicle for providing optimal care. This finding is in line with expert opinion [41] and other research [37], which similarly has found clinicians to perceive trial enrolment as a means to provide their patients with first-class care.

Interestingly, our data showed professionals to be overwhelmingly in (individual) equipoise. We surmise that the nature of TYA-relevant trials may be significant in this. Our data suggest that whilst TYA-relevant trials may involve new treatments, they frequently compare relatively similar regimens, involving differential delivery of familiar treatments. Where differences between treatment arms are subtle, it would seem less likely that professionals might hold strong beliefs regarding the likelihood of particular patients or patient groups benefiting more from one treatment (arm) than another. Arguably, in such cases, the potential for individual uncertainty about involving one's patients in such a trial—and all the concerns and conflicts arising from or fuelled by this—would be reduced.

Although traditional forms of role conflict were largely absent from our data, powerful tensions, or conflicts, were prominent in relation to resource management. Highlighting a backdrop of fiscal crisis, or at least constraint, our interviewees portrayed themselves as having secondary, but nonetheless significant, roles as custodians of scarce resources. They indicated acute awareness of the finite nature of NHS resources and a sense of duty to use these to maximum effect. Many talked at some length of their struggles to reconcile the significant costs associated with involvement in trials with the increasing pressures on their clinical service. Other authors have documented the considerable time and other costs associated with involvement in trials [42–44], with some citing financial constraints as a significant barrier to involvement in (non-commercial) trials [37]. More than a decade ago Snowden et al. [45] argued that financial considerations shaped all aspects of trial work and deserved far closer scrutiny than they had received. Our study helps to fill important gaps in understanding of the actual—and perceived—economics of trials research, and how these mould professionals' decisions and behaviours.

Our data highlight the significant and growing challenges for healthcare professionals of being involved in research—a product of the burgeoning pressure on clinical services and the increasing cost and complexity of clinical trials. These challenges meant that, notwithstanding a deep commitment to trials, only a minority of patients (aged 16+) could be offered treatment through a trial. Scholars working in other jurisdictions have similarly noted that, whilst central to the specialty, in practice, relatively few (adult) oncology patients take part in trials [1, 46]. Our interviewees emphasized their responsibility to make parsimonious choices between trial and other activities and portrayed trials as being in competition both with clinical services and each other. They described making deeply pragmatic decisions about trial engagement, with decisions being oriented towards containing costs and maximising returns. These appeared to follow a crudely utilitarian logic where benefits are calculated according to the number of potential recruits. We see parallels here with Lipsky's depiction of the classic 'street-level bureaucracy', where demands on professionals always exceed the resources available, leading to the development of routines and rationing mechanisms enabling the maximal utilisation of resources whilst maintaining 'a conception of… performance relatively consistent with ideal conceptions of the job' (p.151) [47].

The current system of rewards and penalties for trial activity appeared to reinforce a focus on 'high volume' diseases and to explicitly dis-incentivise more complex or uncertain (in terms of recruitment) trials work. Again, this finding is consistent with Lipsky [47], who argues that discretionary behaviours may 'add up' to policy, but emerge in and from a context which not only structures the choices available, but also provides incentives and sanctions which, in turn, are correctly understood by workers as signalling leadership/management priorities. Such incentivisation schemes are both widespread in the NHS and widely problematized (see for example the debate around the Quality and Outcomes Framework (QOF) [48]). Professionals interviewed for this study recognised that their choices produced patterns—and, as a consequence, inequalities—in access to trials. Interviewees acknowledged that patient groups characterised by small numbers, such as TYA—who tend to present with rare types of cancer—and/or are served by smaller treatment centres, were amongst those most consistently disadvantaged. Differential treatment of different patient groups was, in line with Lipsky, rationalised as serving "the best interest of the greatest number' (p.112) [47]. However, while choices were rationalised, their potential to explain and compound disparities in health outcomes acted as a source of some discomfort to interviewees.

Although the consequences of this new form of role conflict may be similar to those of more traditional forms, the causes are quite different. As such, proposed solutions to traditional forms of role conflict, such as, for example, more or better training on the concepts central to trials and how best to communicate these to

patients [49], would appear to have uncertain relevance. Guidance not on the mechanics of recruitment but instead on how to balance competing obligations and undertake 'boundary work' [50] might be more valuable. Critically, more than a decade ago, Raftery et al. [51] called for better understanding of the organisational and resource aspects of trials and how these impact on research involvement and recruitment. This need does not as yet appear to have been fully met. Recognition of the increasing complexity and cost to health care institutions of hosting trials [41] would seem to us to be a necessary condition of change. Changes to current systems of rewards and penalties for trial activity may also be needed, if health professionals are to be encouraged to make different choices.

Strengths and limitations

Qualitative research can improve understanding of the views, behaviours and decisions of health professionals (and their patients) [28, 52, 53]. It can also prompt the problematization and refinement of concepts and ideas, as demonstrated above. Critiques often concern generalisability; although such generalisability is rarely the goal of qualitative research, it is of interest whether, and with what caveats, findings have wider application. The nature of study samples is key to this. As noted in our Methods, sampling was purposive and continued until we were confident 'saturation' was achieved. Interviewees' views were largely consistent, with strong patterns emerging during analysis. However, importantly, almost two-thirds of our interviewees came from the two largest Scottish health boards. Moreover, not all professionals approached agreed to take part in an interview. Though interviewees' statements regarding their peers and the culture of their specialty suggest that accounts are reflective of a wider, prevailing view, this is not something of which we can be entirely sure. It is possible our sample is skewed towards more research-engaged clinicians; the perspective of those not approached or declining involvement might be different, with the latter perhaps not seeing our study as something to which they could usefully contribute. Also of note is that a relatively small number of nurses were interviewed. Though differences in perspective could not be discerned in our data, future research might usefully explore the perspectives of this staff group in more depth. Looking beyond oncology, Cambrosio et al. [40] advocate caution in extrapolating between specialties and indeed suggest that oncology may be distinctive. We have already highlighted a number of ways in which the trial work the interviewees were engaged in appeared idiosyncratic. We note here, however, that work on trials in other clinical areas, for example, diabetes, has identified similar concerns about financial pressures and difficulties undertaking less profitable studies [16]. Finally, as to whether our findings have relevance beyond the country and health service context in which the research was undertaken, this question hinges on the comparability of service organisation and resourcing. We would encourage readers with direct experience of other healthcare systems to think critically about and explore further the application of our findings to those contexts. Further research may be warranted.

Conclusions

Accounts revealed growing tensions between the ethos of the clinician–researcher, who delivers the best care through the vehicle of trials, and the need to make pragmatic economic choices which inevitably diminish some patient groups' access to such care. The utilitarian logic underpinning professionals' decision-making about engagement in trials was recognised by some interviewees as having consequences for equality of care, in particular, placing patients such as TYA, who typically present with rare types of cancer, and/or who are served by smaller treatment centres, at a disadvantage. The consequences of inequality in access to trials were not explicitly explored in this study. However, denial of those benefits associated by interviewees with trial participation might reasonably be inferred. Moreover, prior research suggests that interviewees' discomfort with the situation has strong foundations. For example, scrutiny of outcomes in TYA cancer patients has indicated that rates of clinical trial participation and improvements in survival (or lack of these) are closely associated [3, 4]. In the United States, 'inclusion in research has come to be seen as an important strategy for reducing health disparities' (p.338) [54], with recognition of inequalities in access to trials along lines of race and gender prompting legislation to promote the involvement of what were subsequently deemed 'special populations' [1]. We do not go so far as to suggest that such an approach be taken here, but argue that at the very least a far franker debate is needed if the issues our study highlights can begin to be resolved.

Abbreviations

NHS: National Health Service; NPT: Normalization Process Theory; PI: Principal Investigator; TYA: teenagers and young adults

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Authors' contributions

AJ conceived the idea for the study, which was then designed and planned by JL and AJ, with input from JH and RH. RH collected and managed the interview data. RH and JL conducted the analysis, with input from NH. RH prepared the first draft of this paper. All authors read and approved the final manuscript.

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Availability of data and materials
The datasets (interview transcripts) reported on in this paper are not publicly available as rendering them entirely unidentifiable would require significant redaction, and the research participants did not consent to public data sharing.

Ethics approval and consent to participate
The study complied fully with the Declaration of Helsinki and was approved by the South East Scotland Research Ethics Committee 01 (REC reference 17/SS/0077). Research governance approval was secured from all participating health boards (13/14 Scottish boards). All interviewees were given written information on the study and provided written consent before the interviews began.

Consent for publication
Not applicable.

Competing interests
The authors declare that they have no competing interests.

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Publisher's Note

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RESEARCH ARTICLE Open Access

The challenges of making informed decisions about treatment and trial participation following a cancer diagnosis: a qualitative study involving adolescents and young adults with cancer and their caregivers

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Abstract

Background: Limited attention has been paid to adolescents and young adults' (AYA's) experiences in the aftermath of a cancer diagnosis, despite this being a time when potentially life changing decisions are made. We explored AYA's and caregivers' experiences of, and views about, making treatment and trial participation decisions following a cancer diagnosis, in order to understand, and help facilitate, informed treatment decision making in this age group.

Methods: Interviews were undertaken with 18 AYA diagnosed, or re diagnosed, with cancer when aged 16–24 years, and 15 parents/caregivers. Analysis focused on the identification and description of explanatory themes.

Results: Most AYA described being extremely unwell by the time of diagnosis and, consequently, experiencing difficulties processing the news. Distress and acceleration in clinical activity following diagnosis could further impede the absorption of treatment relevant information. After referral to a specialist cancer unit, many AYA described quickly transitioning to a calm and pragmatic mind set, and wanting to commence treatment at the earliest opportunity. Most reported seeing information about short term side effects of treatment as having limited relevance to their recovery focused outlook at that time. AYA seldom indicated wanting to make choices about front line treatment, with most preferring to defer decisions to health professionals. Even when charged with decisions about trial participation, AYA reported welcoming a strong health professional steer. Parents/caregivers attempted to compensate for AYA's limited engagement with treatment relevant information. However, in seeking to ensure AYA received the best treatment, these individuals had conflicting priorities and information needs.

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Conclusion: Our study highlights the challenging context in which AYA are confronted with decisions about front line treatment, and reveals how their responses make it hard to ensure their decisions are fully informed. It raises questions about the direct value, to AYA, of approaches that aim to promote decision making by improving understanding and recall of information, though such approaches may be of value to caregivers. In seeking to improve information giving and involvement in treatment related decision making at diagnosis, care should be taken not to delegitimize the preference of many AYA for a directive approach from trusted clinicians.

Background

A growing body of research has looked at the experiences and views of adolescents and young adults (AYA) with cancer. This work has been undertaken to identify challenges and care needs that may be specific to this age group and support development of age-appropriate cancer services [1–7]. When diagnosis has been the focus of AYA research, this work has primarily been undertaken to understand (and help address) delays to diagnosing cancer (e.g. [8, 9]. Limited attention has been paid to AYA's experiences and support needs in the immediate aftermath of diagnosis [10], despite this being the time when critical, and potentially life-changing, decisions about cancer treatments are made. Hence, gaps remain in understanding how best to support decision-making amongst AYA during the time between diagnosis and initiation of front-line treatment.

As commentators have observed, late adolescence and young adulthood is a time when significant physical, emotional and cognitive changes can occur [11]. Hence, AYA may experience different challenges following a cancer diagnosis and have different information and support needs to other age groups [7, 10, 11]. Decision-making may be affected by incomplete development of executive functioning skills, such as planning and impulse control [11]. AYA's decision-making might also be compromised by distress, which may be heightened due to cancer being unexpected in this age group. Consequently, decisions which need to be made very soon after diagnosis, including those about front-line treatment and/or enrolment into clinical trials, may present particular challenges.

Given the varied, complex and – to most of the population – unfamiliar nature of cancer treatments [12–14], newly diagnosed individuals of all ages are often poorly equipped to make treatment-related decisions. Despite being presented with information about the practicalities, risks and benefits of proposed treatment(s), research suggests patients (in general) often struggle to understand fully important issues such as treatment intent and prognoses [12]. Where, as might be the case in a clinical trial, information encompasses two or more distinct regimens, cognitive demands may be further amplified.

AYA with cancer have lower improvements in survival rates than children and older adults and their low participation in cancer trials is believed to be a contributory factor [15–17]. This has prompted calls to identify and address barriers to trial enrolment in this age group [7, 11]. Specifically, commentators have highlighted the need to investigate AYA's psychological response to a cancer diagnosis [18] and understand the challenges to enrolling individuals to clinical trials at this time [16, 18]. The importance of consulting caregivers and exploring their perspectives has also been noted, as these individuals may influence AYA's decision-making [19].

Some research has now explored barriers to clinical trial participation from AYA's perspectives. Studies have, however, been limited by questionnaire designs [20, 21], single site recruitment [22, 23] and/or because they have focused on individuals with one type of cancer [20, 24, 25]. Participant samples have also tended to be skewed towards adolescents under the legal age for independent decision-making and consent. Hence, it is unsurprising that some studies have found parents to be central drivers in decision-making [21, 22]. Due to these limitations, a recent systematic review concluded that AYA's perceptions and attitudes towards clinical trial participation remain under-explored, especially amongst those treated in adult cancer centres [11]. In addition, it is notable that research and debates about improving trial participation by AYA have focused on how to increase enrolment, rather than the quality of their decision-making.

Here, we report findings from interviews undertaken with AYA diagnosed with cancer whilst aged 16–24 years and their caregivers. The study was conducted in Scotland, UK, where individuals aged 16+ years have the legal right to make decisions about medical procedures and treatment, including trial participation. We explored interviewees' experiences of, and views about, making decisions about treatment and/or trial participation following a cancer diagnosis, in order to understand, and help facilitate, informed treatment-related decision-making in this age group.

Methods

We describe our methods, below, in line with the consolidated criteria for reporting qualitative studies (COREQ) [26]. Qualitative methods are recommended when little is known about the area under investigation,

as they allow findings to emerge from the data, rather than testing pre-determined hypotheses [27, 28]. We employed an inductive, semi-structured interview design entailing simultaneous data collection and analysis. This afforded participants the flexibility to raise issues they perceived as salient and allowed issues identified in early interviews, including those unforeseen at the outset, to inform the questions asked, and areas explored, in later ones.

Theoretical framework(s)

Our work took its epistemological orientation from critical realism, a philosophy which, in simple terms, treats accounts as indicative of participants' lived experiences and perceived realities, whilst recognising that what is disclosed in interviews is contextually-mediated and influenced by a variety of social, circumstantial and other factors [29, 30]. Our methodological orientation was qualitative description [31, 32], a pragmatic approach focused on the identification and description of minimally-theorised explanatory themes. Hence, we did not embark on data collection with allegiance to any particular theoretical concepts; however, literature on experiences of diagnosis sensitised us to the possibility that the events leading up to a (cancer) diagnosis might provide a context for, and influence and inform, individuals' emotional reactions and subsequent treatment decision-making [33, 34].

Context/setting

Our study was conducted in Scotland, UK. National Health Service (NHS) Scotland delivers cancer care, free of charge, through 14 health boards and more than 20 hospitals. Most patients diagnosed with cancer at age 16 or above receive treatment in an adult hospital, though paediatric hospitals deliver care to some patients aged 16–19 years. Specialist AYA units are available within the four largest health boards.

Sampling and recruitment

Around 180 young people aged 16–24 are diagnosed with cancer in Scotland each year. From this pool of potential participants, we sought, purposively, to recruit AYA with varying characteristics in terms of age, gender, diagnosis, place of care, and trial experience. Clinical teams facilitated the research by identifying AYA diagnosed, or re-diagnosed, with cancer when aged 16–24 years from three paediatric and five adult cancer centres in Scotland. Initial contact with AYA patients was made by members of their direct care team, who offered these AYA recruitment packs containing an opt-in form returnable to the qualitative research team. Due to the range of direct care colleagues involved in this process we cannot say definitively how many AYA were approached, or how many chose not to take part.

Where opt-in forms were returned, contact was made by the project researcher (RIH) who talked through the details and practicalities of taking part in the research. AYA who agreed to take part in an interview were asked to give a recruitment pack to a caregiver (e.g., a parent or partner) or another individual (e.g., a friend) who had been influential in their decision-making about cancer treatment and/or care. Again, these packs included opt-in forms, on receipt of which contact was made by RIH. All AYA were recruited between November 2017 and December 2018, whilst caregivers were recruited between February 2018 and January 2019.

Data collection

Interviews were undertaken by RIH, an experienced (non-clinical) qualitative researcher, at a time and location convenient to participants – typically their own homes or the hospital at which AYA were receiving care. In both such locations, interruptions by family members and/or healthcare professionals were relatively common. In such situations, interviews were generally paused, and resumed once privacy was restored. While AYA and caregivers were invited to take part in separate interviews, four AYA elected to be interviewed with one or more parent-caregiver. Topic guides helped ensure the discussion remained relevant to the study aims, while affording scope and flexibility for participants to raise issues they perceived as salient, including those unforeseen at the outset. Key areas explored relevant to the reporting in this article are outlined in an Additional file 1: (a Microsoft Word document, with the file extension .docx). In developing topic guides we drew on the expertise of clinical co-investigators and AYA advisors, as well as relevant literature. Topic guides were revised in light of emerging findings and adapted in situ, i.e. used flexibly, to take account of variations in age, development, education and interviewees' emotional states, and to probe and explore in more depthissues which particular participants chose to disclose. Whilst our goal was to elicit rich information, we were cognisant of the responsibility not to cause avoidable/unnecessary distress. Hence decisions were made in some instances not to pursue certain issues in depth. Interviews typically lasted 1–2 h; all were digitally-recorded and transcribed, with interviewees' consent. Information on the context and non-verbal components of the interview were recorded in field notes. Data collection continued until our sampling ambitions had been broadly satisfied and no new findings were identified in new data collected (data saturation).

Data analysis

Two highly experienced, non-clinical qualitative researchers (RIH and JL) analysed the data, using the method of constant comparison [35] to identify key

themes in AYA and caregiver accounts. Both researchers immersed themselves in the data and read interviews through repeatedly, before independently undertaking preliminary analyses. They wrote separate reports and then met to discuss their interpretations and agree on a coding frame which captured key themes. Coded datasets were subjected to further analyses to allow more nuanced interpretations of the data and identification of illustrative quotations, with a qualitative data-indexing package, NVivo (Version 11, QSR International Pty Ltd., Doncaster, Victoria, Australia), used to facilitate data coding and retrieval. Emerging findings, supported by illustrative quotations, were shared with the wider research team, members of the study advisory groups, and participating AYA and their peers (via a workshop convened in the final phase of the project). Feedback was largely confirmatory and was used to inform study recommendations.

Results

18 AYA were interviewed, of these 13 nominated one or more caregiver who was also interviewed (11 mothers, 3 fathers and 1 partner). The remainder either did not wish to involve a caregiver or felt unable to do so (e.g. as their family resided in another country and/or spoke limited English). See Table 1 for more details about the sample. As participants were drawn from a very small population and are, therefore, potentially easy to identify, we have only been able to provide limited clinical and personal information in our reporting, below.

Below, we begin by describing AYA's reactions to diagnosis and their experiences in its immediate aftermath, in order to illuminate the context in which they were confronted with decisions about their treatment. Building on this, we document AYA's responses to treatment-relevant information at this time, and identify reasons why these individuals did not choose, or push, to be actively involved in decision-making. We then consider how caregivers sought to play support(ive) roles to help compensate for AYA's lack of engagement with information and ensure AYA's clinical interests were met. Key themes which structure our reporting include: difficulties processing the news; a rush of emotion; a whirlwind of activity; struggling to absorb information; going into a (recovery-focused) zone; disengaging from challenging information; detaching from decision-making; seeking to protect and support AYA; and conflicting information needs.

Difficulties processing the news

AYA and caregiver participants described a variety of circumstances and events which had led to a cancer diagnosis and referral to a specialist cancer unit. Most reported having been made aware that the results of key tests (such as MRI scans and biopsies) indicated that they/AYA might or did have cancer by a health professional other than an oncologist (e.g. a surgeon). Many described themselves/AYA as having experienced marked deteriorations in health by this point. Some AYA commented that they had felt so unwell, overwhelmed, and exhausted that they had struggled initially to process the news. A16, for example, a young man eventually diagnosed with osteosarcoma, described having been in "unbearable pain" when cancer was first broached, and how, as a consequence, "I didn't process the information… I wasn't absorbing anything." Others described how the after-effects of a general anaesthetic and/or initiation of pain relief had interfered with their ability to process what they were being told. This included A14, a young man diagnosed with a form of sarcoma uncommon in young adults. A14 reported how, when first advised that his symptoms were suggestive of this rare cancer, he had been "so dosed up on morphine that I had no idea what that [diagnosis] meant."

A rush of emotion

In the majority of cases, shock and extreme distress were described as AYA's over-riding reactions to the news that they might, or did, have cancer. As the mother of a young man diagnosed with a haematological malignancy recalled: "the first thing (A09) said to me, he said, am I going to die? And I just remember tears pouring down his face, so he did get a big fright." Notably, however, some AYA participants also described having felt relief, due to the severity of their symptoms and the prospect of now receiving the correct treatment. This included A05, a young woman ultimately diagnosed with bone cancer, who reported presenting to her GP and other professionals on repeated occasions before finally being diagnosed. This young woman described having eventually gone to an accident and emergency department (A&E) in a state of desperation and extreme pain: "I was just in agony. I didn't sleep, couldn't eat". Hence when a series of tests revealed she had a tumour, she described experiencing a range of conflicting emotions:

> "I'm not going to lie… I was a wee bit stunned and then I was really upset, we were all crying and then I was a bit like, what's going to happen… but then part of me, in a weird way was like, this is gonna take the pain away. Like the pain was that bad."

A whirlwind of activity

In many cases, the sudden acceleration in activity which followed diagnosis left little time for reflection, and presented additional challenges to AYA processing the news. Many participants reported how they/AYA were

Table 1 Characteristics of study participants

AYA with cancer ($n = 18$)		
Age: median (range)	At diagnosis	19 (16 24) years
	At interview	20 (17 26) years
Gender, male: n (%)		14 (78%)
Ethnicity: n (%)	White British	14 (78%)
	Non white British	2 (11%)
	White non British	2 (11%)
Education/employment at diagnosis: n (%)	Employment / work based training	6 (33%)
	School / college	6 (33%)
	Undergraduate studies	3 (17%)
	Not in education / employment	3 (17%)
Diagnosis: n (%)[a]	Bone sarcoma	6 (33%)
	Leukaemia	4 (22%)
	Germ cell tumour	3 (17%)
	Other sarcoma	2 (11%)
	Lymphoma	1 (6%)
	CNS tumour	1 (6%)
	Melanoma	1 (6%)
Diagnostic type: n (%)	Primary cancer	16 (89%)
	Relapsed cancer	2 (11%)
Place of care: n (%)[a]	Adult hospital with AYA unit	14 (78%)
	Paediatric hospital with AYA unit	3 (17%)
	Adult hospital without AYA unit	1 (6%)
Reported enrolment in a trial: n (%)		5 (28%)
Interviewed independently of caregiver(s): n (%)		14 (78%)
Caregivers ($n = 15$)		
Relationship to AYA: n (%)	Mother	11 (73%)
	Father	3 (20%)
	Partner	1 (7%)
Ethnicity: n (%)	White British	14 (93%)
	Non white British	1 (7%)
Own occupation at AYA's diagnosis: n (%)	Professional	8 (53%)
	Semi professional / Skilled	6 (40%)
	Unskilled	1 (7%)

[a]Percentages do not sum to 100% due to rounding

sent straight onto a specialist cancer unit, sometimes within a matter of hours of receiving their initial results. As A05's father recounted in a joint interview with his daughter:

> "things just moved so quickly... it was like a whirlwind. It just seemed to go from him telling us, and us trying to take it in, and then we had to get to (Children's Hospital) to meet with (consultant oncologist) and then things just went very, very quickly."

Participants also reported a further escalation of activity following arrival at the cancer unit, wherein, after an initial consultation, most underwent a battery of tests and medical procedures in quick succession to confirm (the type of) cancer and/or establish its spread before treatment could be determined and initiated:

> "after I saw my (Consultant Oncologist) for the first time, he arranged loads of different scans Because it was very common for it to, have spread elsewhere in the body, particularly the lungs, and possibly the

bones. Obviously that was extremely harrowing. I got a CT scan, I got a bone scan, I got a full body scan, heart scan, kidney scan." (A04)

Struggling to absorb information

AYA participants were generally able to recall some elements of the consultation in the cancer unit where test results and treatment plans were first discussed. However, many noted how, due to extreme exhaustion, the shock of diagnosis, and the limited time they had had to process the news, they had struggled to absorb and engage with treatment-relevant information. A18, for instance, described a meeting with her oncologist shortly after surgery to remove a large pelvic mass: "We were obviously talking about treatment (radiotherapy). But I'd just come out from surgery, so I was too tired". This situation was confirmed by her mother who noted how:

"she was exhausted and I think a lot of the time (daughter) was leaning on me zonked out … she didn't really, really understand the full implications of what it [treatment] was going to entail."

Going into a (recovery-focused) zone

Notwithstanding their harrowing experiences around the time of diagnosis, most AYA participants described transitioning, relatively quickly, from a mind-set of shock, extreme distress and fear, to one which had been calm, largely devoid of emotion, and deeply pragmatic. This included A12, a young man diagnosed with acute lymphoblastic leukaemia, who noted how, when a consultant discussed his diagnosis and its implications:

"Mum and dad started crying... but I didn't, I just sat on my bed and was quite matter of fact about it… I'd had a little cry earlier, when I'd seen the (Teenage Cancer Ward) sign or whatever, but I'd kind of accepted my fate, already."

A03, a young woman with Ewing's sarcoma, likewise described having entered a "zone" following her arrival at the cancer unit, wherein her focus had rapidly shifted from shock and distress to recovery, and commencing treatment at the earliest opportunity to achieve this:

"you kind of just go into like a zone, you're like, it is what it is … you know, you need to get on with it, either way, so. he sat and he explained everything [test results] to me, and I was just like right, I just wanted to get on with it, just let's go, I wasn't like upset or anything like that… probably when I'm finished I will be like, oh my God, how the hell did I do that? But when you're in that zone, you're like right, this is it, I just need to focus on getting better."

Some participants noted how this mind-set had been fostered and enabled by the speed with which investigations had been undertaken and results had come through. Indeed, several individuals described having actively welcomed this forward momentum and accelerated activity, precisely because it had left little time to worry about their prognosis and fixate on negative scenarios:

"I've been lucky in the sense that. it's just been literally full steam ahead … and I've preferred it like that because you've not got any time to think about things because it's just like, right you've been diagnosed, we're starting treatment we can start next week … it doesn't give you time to think, like 'oh no, this is what I've got, like, what's going to happen to me?' You know, all that stuff that's negative." (A16)

Positive steers from health professionals were also described as having been welcomed and as having had a galvanising effect:

"It was all really positive stuff, it was never anything really negative at all actually which suited me… From day one, he said to me, going to get you better, going to beat this kind of thing… So, I just wanted to get started then and there." (A16)

Disengaging from challenging information

In light of their focus on recovery and attendant wish to get on with treatment, most participants reported seeing information about short-term side-effects of treatment (e.g. hair loss, nausea) as having had limited relevance to their thinking and priorities at the time. A14, for instance, a young man diagnosed between leaving school and starting university, reported how he had not "really given a damn about short-term side-effects, the things that do matter to me are the long-term side-effects, what's my life going to look like in five years, 10 years."

Some AYA participants also described having chosen to disengage from potentially distressing information, despite its potential relevance to treatment decision-making, due to its potentially detrimental emotional impact. For example, A18, the young woman whose early experiences were described above, remarked: "like chemo and that… I was just too sad to read them [leaflets]… Yeah just, I'd just end up crying". This response appeared especially marked in relation to information about prognosis. Some AYA described themselves as having had no desire for detailed information on their prospects, and appeared keen to prevent or circumvent negative thinking, as A16 explained:

> "he (consultant oncologist) never told me what stage of cancer I was in, I never asked, he never mentioned once survival rates, anything like that, which I was happy with."

Detaching from decision-making

Though many decisions had to be made about AYA's treatment, the majority of AYA participants saw themselves as having had limited opportunities for involvement. As one young man described, "decision-wise… it was all laid out for me in a way, there wasn't really a lot of room for negotiations" (A15). A similar experience was recounted by A14:

> "I wasn't presented with much of a decision when I saw (oncologist)… He explained that he wanted to treat me under a regimen which was developed as part of a clinical trial… He basically told me that you'll have a longer duration of treatment, but it substantially increases your chances of a cure and I was like, fine."

None of these individuals questioned or challenged this directive approach in retrospect. To the contrary, AYA participants often described having preferred to defer decision-making to health professionals, due to having felt unwell and overwhelmed, and just wanting to get on with treatment. A particularly poignant example was provided by A17, a young man diagnosed with a brain tumour following an emergency admission after collapsing at home. Having undergone surgery to remove and biopsy the tumour, this individual described his state of exhaustion as extreme: "I was sleeping after it for about 20, 22 hours per day." He also described having felt "gutted" on discovering how advanced his brain tumour was (stage IV). A17 made it clear that, at the time, he had just wanted to be looked after and for trusted specialist health professionals to make decisions for him: "do what's best for me, that's what I would say."

Enrolling without fully understanding

When AYA participants had been invited to take part in a clinical trial following diagnosis, more concrete and tangible decisions had needed to be made. As those participants ($n = 5$) who recalled such an approach as having taken place reported, discussions about treatment and trial participation had taken place concurrently. This was because decisions about trial participation had needed to be made rapidly, before treatment could commence. In keeping with the above accounts, these individuals described how trial-participation decision-making had taken little account of information and had involved limited deliberation. The following interviewee, for example, one of the youngest in our sample, reflected on how:

> "It was more like a gut [reaction], I mean, I did ask (Mum) and (Dad) what they were thinking, but I didn't really care, it was more just, it's probably the right thing to do. So, I just did it." (A07)

Indeed, rather than engaging with trial-related information, participants described taking cognitive shortcuts and basing their decision on the recommendation of, or a strong steer from, the consultant who recruited them:

> "I didn't really think about it properly, because… the main factor was actually that, I trusted Dr (Name), and, he seemed keen on it, and he seemed to be wanting to persuade me to do it, so, I was quite happy to take part, without maybe knowing fully, what it entailed." (A12)

Some noted how, as a consequence, their understanding of the trials into which they had been recruited had been very limited, as A01 explained:

> "it was all a bit of a blur at that point … I didn't quite have an understanding of the trial. To be honest I don't quite have an understanding of it now."

None of these individuals expressed regret about trial enrolment, and all described feeling that their health professionals had acted in their best clinical interests. However, some AYA did question in hindsight whether they had made fully informed decisions about taking part. As these individuals suggested, the problem had not resulted from the information they had been given at trial enrolment but, rather, the context and timing of the approach:

> "Maybe, the bombarding of information in the first week. It's a lot of information to take in, and then to start treatment so quickly…. Erm… so yeah like, to spread it out over a longer period of time, so that people have that time to process all of it." (A01)

Caregivers: seeking to protect and support AYA

Caregivers, who were mostly parents, described having wanted to do everything they could to protect and support AYA patients following a cancer diagnosis. Many

reported concerns that – as AYA themselves had suggested – their son/daughter had been unable or unwilling to engage with information and actively participate in consultations at the time treatment plans were discussed. This included the mother of one young man (A10), who had been in employment and preparing to buy his first house at the time of diagnosis. M10 observed how her son, despite leading a relatively independent and adult life:

> "wasn't well, he wasn't taking part in the conversation, it was probably more me. And I think he was just exhausted. And obviously a bit, well, scared."

Likewise, A12's mother noted how her son had been, "quite unwell really, (so) even like concentrating on reading documents (was) hard for him." Reflecting further, she added: "at times I felt that (son) didn't have the questions to ask, because (son) wasn't well enough to be asking questions."

Other parents described how, because of their worries that AYA had been unable to concentrate and had not wanted to deliberate at length over decisions, they had found it very challenging that it had been the young person, rather than themselves, to whom treatment-related information had been cascaded, and, moreover, who had been charged with responsibility for decision-making. As the mother of A07, the 16-year-old whose "gut" decision to participate in a trial was described earlier, explained:

> "ultimately the decision, on the trial, was (son's). I struggled with that… I struggled with the fact that (Consultant) initially was talking to (son), because he can give consent, he doesn't need our input at all. But I didn't feel (son), after being told that overwhelming news, was kind of, you know, he needed support to help him make the decisions." (M07)

Acting as retainers, investigators, sounding-boards and influencers

In response, parents/caregivers described having undertaken various overlapping support roles to help ensure AYA's clinical interests were met. Specifically, they described having made efforts to be present in consultations to assimilate information on AYA's behalf; for instance, by listening carefully and taking detailed notes. These individuals also described how, following consultations, they had gone away and carefully read all the written materials which AYA were given, in order to come back and ask targeted and focused questions on their behalf.

Most parents/caregivers also reported having undertaken their own investigations before any treatment or trial participation decisions were finalised. This included researching the oncologist's credentials, finding out more about cancer and potential treatments, and/or researching trials online (when these were offered to AYA). In some cases, parents/caregivers had sought advice from personal contacts who had specialist cancer knowledge. Parents/caregivers described having undertaken these investigatory roles to help ensure AYA received the best care from the most qualified individuals, and to lobby for changes if necessary:

> "Because we had a few days before we met the oncologist, I was able sort of to have my own questions as to what they were gonna do, and how they were gonna do it, and were they the best?" (M06)

Caregivers also noted how, by virtue of having undertaken their own research, they had been better placed to act as sounding boards before decisions about treatment/trial participation were finalised and consent forms signed. To this end, they also noted how they had been well situated to nudge, sway and/or endorse decisions as necessary. A01's mother for instance, described having encouraged and supported her daughter's decision to take part in a trial after carefully reading all of the documentation and being reassured that her treatment and care would not be adversely affected:

> "when you sat down and got the information, it was only the method of treatment that was differing, it didn't affect the outcome and that was the most important thing to me… so… I said, well you know, it's up to you at the end of the day, it's your body, I said, but I think it's a good idea. So [my role in the decision-making process] was supportive." (M01)

While these caregivers generally endorsed (trial participation) decision-making, they also noted that if they had had any concerns, they would have attempted to nudge AYA into making a different decision. As A07's father described:

> "We did have a friend who's an oncologist… She'd gone away and done her own research, came back and said, It is a good trial… it's probably a good one to go on – if she'd come back and said something different, we might have tried to talk (son) out of it." (D07)

Conflicting information needs

In undertaking these support(ive) roles, parents/caregivers recognised that there were potential conflicts between their own information needs and those of AYA. While AYA wanted to maintain a positive, recovery-

focused outlook, parents/caregivers described needing realistic information, including information about AYA's prognosis, to help ensure the best decisions were made:

> "it might be that some doctors are more upfront about it, but Dr (Name)... was wholly positive with his demeanour, and I actually asked him, 'cause I didn't want to ask him in front of (son), I asked him, I pulled him outside that first day, and I said, with (son's) permission I asked him, You know, is, could he die from this?" (M12)

As parents further noted, this could sometimes mean treading a delicate line between satiating their own needs and requesting information which could cause distress to their child. A particularly poignant example was provided by M06, whose son had been diagnosed with an extremely rare form of cancer. M06 had been very anxious to ensure her son received his care from health professionals with prior expertise of treating his kind of cancer, even if this meant moving to another cancer unit. To do this, she had found it necessary to ask very difficult questions (e.g. about survival rates) which, as she realised, could cause her son upset:

> "Because they did say they haven't had anybody like him, which was a bit alarming, because I felt, well, how do they know how to treat him… I asked, well why isn't he going to (City) where obviously they have got more experience with that type of tumour… I asked was there any place in Scotland where there'd been more patients that had had it? And what was the outcome? But it's hard saying that when he's sitting there… You don't want to say in front of them: What's the success rate? And you don't want to put any more pressure on (son) by asking too many things."

Discussion

This study has highlighted the profoundly challenging context in which AYA diagnosed with cancer find themselves confronted with decisions about front-line treatment and/or trial participation. In keeping with other studies [21, 22, 24], we have shown how the physical effects of cancer and the shock and distress of diagnosis can influence AYA's initial response to diagnosis, and hamper their ability to process difficult news at this time. In addition, our study has highlighted how intense emotion, and the escalation of clinical activity that follows diagnosis, can further impede AYA's ability to absorb and process important (treatment-relevant) information. Notably, we have drawn attention to how AYA may quickly transition from a state of shock and distress to a mind-set focused on survival and recovery, which our participants described as "going into a zone". Whilst in the zone, AYA described wanting and valuing opportunities to filter out negative scenarios. Although this mind-set may act as an important coping strategy in the aftermath of a cancer diagnosis, we have shown how it can further compromise engagement with treatment-relevant information, including information about prognosis and treatment side-effects, and thereby undermine informed decision-making. In general, we found AYA's interest in engaging in decision-making about front-line treatment to be low, with most indicating a clear preference for a strong professional steer at this time. Caregivers expressed concerns about the quality of AYA's decision-making, and described attempting to compensate for their limited engagement with relevant information. However, in seeking to support AYA, and help ensure that they received the best treatment/care, these individuals could have conflicting priorities and information needs.

As our findings suggest, professionals tasked with facilitating AYA's involvement in decision-making about (front-line) treatment and/or trial participation are often confronted by major challenges in the form of AYA's physical and emotional states at and in the immediate aftermath of a cancer diagnosis. Such challenges have led commentators to question whether it is really possible to obtain informed decisions about, and consent to, front-line treatment and trials at this time. With regard to trials, some have even asked whether it is (ethically) appropriate to attempt to recruit patients so soon after diagnosis, and where decisions about participation need to be made rapidly, so that treatment might commence [16, 22–24]. This issue of how to achieve informed decisions and/or consent under pressure is of considerable wider interest, with challenges highlighted in a range of studies. This includes work involving parents of younger children diagnosed with cancer, where, again, treatment often needs to be initiated very soon after diagnosis [36]. Approached about trial participation within hours or days of learning their child had cancer, these parents similarly struggled to assimilate and reflect upon (complex) information needed to make informed decisions. As with our AYA participants, this was due to upset, shock, and the limited timeframes available [36]. Similar concerns have also been reported in studies exploring the issues arising when recruiting individuals into (non-cancer) clinical trials in other acute/emergency situations, where again distress may be heightened and decisions need to be made quickly [37–39].

The literature offers some pointers as to potential ways of overcoming the challenges presented by poor health, distress, and short timeframes. However, while some of these proposals have been recommended for use in AYA and/or paediatric populations newly diagnosed with

cancer, they are, as yet, of unproven efficacy. These proposals include using novel communication strategies (e.g. audio or video platforms), decision-aids and question prompt lists [11, 17, 40–43]. It has also been suggested that information could be given out in smaller quantities, over varied periods, and that investigators should systematically ask individuals to recall the information given when decisions are confirmed, and consent taken [36]. It is noteworthy that all such approaches focus on improving understanding and recall of information (e.g. about the risks and benefits of treatment and/or trial participation). Given our own findings that AYA may be too unwell to assimilate and recall information needed to make fully informed decisions, and may be unwilling to engage with negatative scenarios (e.g. about prognosis and treatment side-effects), these approaches may be of limited value for AYA patients confronted with decisions at diagnosis. Moreover, because of the expediency with which their cancer treatment often needs to commence, approaches that require information to be delivered at several time points may be less feasible than for other patient groups presenting with less acute forms of disease.

AYA participating in our study described how, following diagnosis, there had seldom appeared to have been any major treatment-related decisions for them to make. Agreeing to (or declining) the course of action proposed by professionals was not typically viewed as a real choice/decision. This finding is perhaps unsurprising, given that observational work undertaken in cancer multi-disciplinary team meetings has demonstrated that health professionals tend to reach a consensus about which treatment is best before delivering this recommendation to the patient [44]. Other research has indicated that, in oncology, it is common practice for clinicians to make explicit recommendations, and for there to be less scope for negotiation of treatment plans than in other clinical specialisms [45]. Hence, shared decision-making in oncology, especially in situations where professionals believe there is a course of action which is in a patient's best clinical interests, has been reported to be rare [46]. Notably, none of the AYA in our study questioned, in hindsight, the validity and acceptability of the directive approach they described; to the contrary, these individuals gave little indication of wanting treatment choice at this time. Whilst, as others have suggested, this preference might be due to AYA having not yet fully developed executive functioning skills [1], our findings suggest more complex multi-factorial explanations, in which disease acuity and the limited time available to make decisions play important roles. Indeed, it is relevant to note that amongst other patient groups where quick treatment decisions have also needed to be made, similar preferences have been highlighted to those reported by AYA. For example, in a study involving adults with haematological malignancies who were in life-threatening situations and under extreme emotional strain, it was found that these individuals similarly leaned towards directive approaches [47]. Some commentators have argued for greater recognition of the legitimacy of this preference for a directive rather than a shared decision-making approach [48]. Mol [49] problematizes the common framing of choice as the ideal, positing that choice does not necessarily result in good care, and may leave patients feeling burdened and/or with an unhelpful illusion of control. Even key proponents of shared decision-making have acknowledged that such an approach may not be appropriate or feasible in all decisional contexts, and in some a more directive (paternalistic) approach may be both preferred and required [50, 51].

While a directive approach may be acceptable when health professionals are clear about the most efficacious and acceptable form of treatment, it is potentially more controversial in the context of clinical trial recruitment, where, in principle, equipoise exists and there is uncertainty with regard to which type of treatment is in an individual's best clinical interests. In these situations, it may be problematic to involve consultants in trial recruitment, given that AYA tend to base their decisions upon trust in those individuals, as opposed to on careful engagement with trial-relevant information. It may be even more problematic still to follow the recommendation, made by others, that those staff to spend as much time as possible with AYA to develop rapport, in order to improve trial recruitment [16, 24]. Rather, consideration could be given to using more neutral parties in the information-giving and consent process for trials [36]. Input from psychological services might also be considered to help reduce distress following a cancer diagnosis, and, through this, potentially to increase decisional involvement by AYA [40].

The role of caregivers in such situations is both interesting and potentially important. Our study extends understanding of parent-caregivers' concerns, and the work they undertake to support AYA. Previous work involving parents of younger children has shown that these individuals often adopt advocacy and investigator roles to help ensure the best decisions are made for their child [19, 52, 53]. Our study demonstrates that, once "children" reach the legal age to make their own decisions about treatment, parents often continue to play supportive roles. These include: attending consultations; asking questions on AYA's behalf; undertaking research; acting as sounding boards; and, "nudging" AYA towards (different) decisions. Future research could explore the best ways of engaging this support, while being sensitive to the fact that parents' priorities and information needs may conflict with those of AYA. Any such work must further recognize that,

through choice or circumstance, not all AYA have parental support following a cancer diagnosis.

Strengths and limitations

Using a flexible and open-ended approach, we have been able to bring new and important insights to the literature. The inclusion of a caregiver alongside an AYA perspective has revealed a more complex decision-making dynamic than erstwhile recognized. In the context of joint interviews, AYA and caregivers operated as co-producers of knowledge, with caregivers helping AYA to fill gaps in recall arising from acute illness and distress. However, in some instances, this might have inhibited open discussion about the difficulties and worries experienced (by AYA or caregivers). While we attempted to achieve heterogeneity in our AYA sample, we interviewed more males than females. Some of the stoical reactions reported may therefore reflect cultural and gendered norms and expectations regarding masculinity. Because participants' accounts were retrospective, they may have been subject to recall bias. Hence, future prospective (longitudinal) research could be considered, including observation of consultations where key discussions about treatment/trial participation take place.

Conclusions

Treatment in the field of AYA cancer care is guided by complex and evolving protocols, often covering long periods of time. Hence, even under optimal conditions, the cognitive demands of absorbing, processing and employing treatment information for the purposes of decision-making are substantial. Our findings indicate that, due to the context in which AYA are confronted with decisions about front-line treatment, including, where available, treatment through a trial, their decision-making may not be fully informed. The survival/recovery-focused mind-set AYA may adopt as a coping strategy further runs counter to meaningful engagement with information and decision-making. Hence when AYA are first diagnosed with cancer, rather than delving into the detail of the treatment and/or trial, they may prefer to make decisions directed by a trusted clinician. Though this preference may be viewed/interpreted differently on account of their age, there is ample evidence to suggest that AYA are far from unique in favouring a directive approach. Care should be taken not to delegitimize this preference/choice in the process of developing and implementing strategies to improve information-giving and encourage meaningful involvement in decision-making.

Supplementary information

Supplementary information accompanies this paper at https://doi.org/10.1186/s12913 019 4851 1.

Additional file 1. Key areas explored in the AYA and caregiver interviews.

Abbreviations

A&E: Accident and Emergency Department; AYA: Adolescent and Young Adult; NHS: National Health Service; UK: United Kingdom

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Authors' contributions

JL, ABJ and JH designed the study. RIH collected data, and JL and RIH undertook data analysis. JL drafted the manuscript, with assistance from RIH. DAC, FJC, JH, NH, DR and ABJ reviewed and edited the manuscript. RIH revised the manuscript, in response to reviewer comments. All authors read and approved the final manuscript.

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Availability of data and materials

The datasets (generated and/or reported in this paper are not publicly available due to the very small number of AYA diagnosed with cancer in Scotland each year. This means that it may be possible even after the removal of obvious identifiers (e.g. names and locations) for some individuals, such as health professionals providing relevant services, to identify the young people who took part. These datasets are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

Ethics approval was secured from the South East Scotland Research Ethics Committee 01 (REC reference 17/SS/0077). All interviewees were provided with written information about the study and gave written consent prior to participation.

Consent for publication

All interviewees consented to the use of quotations from their interviews in publications arising from the study.

Competing interests

The authors declare that they have no competing interests.

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ORIGINAL RESEARCH PAPER

WILEY

Adolescents and young adults' (AYA) views on their cancer knowledge prior to diagnosis: Findings from a qualitative study involving AYA receiving cancer care

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Abstract

Background: Cancer is rare amongst adolescents and young adults (AYA). Previous research has reported (healthy) AYA's knowledge of risk factors and symptoms as limited, with this potentially leading to delays in help-seeking and diagnosis.

Objectives: We explored AYA's views on their cancer knowledge prior to diagnosis and if/how they perceived this as having affected their experiences of diagnosis and care.

Methods: We interviewed 18 AYA diagnosed with cancer (aged 16-24 years). Interviews were recorded and transcribed verbatim. We undertook qualitative descriptive analysis, exploring both a priori topics and emergent themes, including cancer knowledge prior to diagnosis.

Results: Adolescents and young adults characterized their knowledge of cancer and treatment prior to diagnosis and treatment initiation as limited and superficial. AYA perceived gaps in their knowledge as having profound consequences throughout their cancer journey. These included: hindering recognition of symptoms, thereby delaying help-seeking; impeding understanding of the significance of tests and referrals; amplifying uncertainty on diagnosis; and affording poor preparation for the harsh realities of treatment.

Conclusions: Adolescents and young adults perceived their limited cancer knowledge prior to diagnosis as affecting experiences of diagnosis and initial/front-line care. These findings prompt consideration of whether, when and how, AYA's knowledge of cancer might be improved. Two broad approaches are discussed: universal

Angela B. Jesudason and Julia Lawton are joint last authors.

education on AYA cancer and/or health; and targeted education (enhanced information and counselling) at and after diagnosis.

Patient or Public Contribution: Our work was informed throughout by discussions with an advisory group, whose membership included AYA treated for cancer.

KEYWORDS

adolescents, cancer, information, knowledge, patient care, qualitative research, young adults

1 | INTRODUCTION

Cancer is rare amongst adolescents and young adults (AYA), but its incidence is increasing.[1] Its impact on young lives can be profound. A growing body of quantitative and qualitative work has considered AYA's experiences of cancer, diagnosis and treatment/care.[2-13] That work suggests the experiences and needs of this age group differ both from those of older adults, who comprise the majority of cancer patients/service users, and of younger children.

We recently completed a study which explored AYA's experiences of decision-making about front-line cancer treatment and their perspectives on participation in (Phase 3) clinical trials. That study highlighted significant challenges to informed decision-making about both treatment and trials, including difficulties engaging with (disease and treatment-related) information in the immediate aftermath of diagnosis.[14] The early stages of our analysis revealed an additional, important theme: knowledge of cancer and its treatment *prior* to diagnosis. Identification of this theme prompted further, more detailed analytical work, which we report in this paper.

Previous research on AYA's cancer knowledge provides a context for this analysis. That research has shown that (healthy) AYA have limited knowledge of key cancer risk factors and symptoms.[15-18] Studies have also highlighted deficiencies in (healthy) AYA's knowledge of cancer-related personal health surveillance (self-examination) practices and public health initiatives (such as HPV vaccination and cervical screening programmes).[19-25] It has been suggested that these knowledge gaps may lead to delays in symptom recognition, help-seeking and diagnosis. Late diagnosis is commonly associated with poorer outcomes, and preventable delays are a prominent concern in the AYA cancer literature.[2,5,26]

However, prior research has focussed on understanding (typically measuring) ***healthy*** AYA's knowledge of cancer. Very limited attention has been given to the views of AYA diagnosed with cancer about their pre-diagnostic cancer knowledge and, whether and, how, they perceive this as having influenced their experiences of diagnosis and care. Understanding knowledge is important, as studies exploring the information (and other) needs of AYA with cancer further from diagnosis and/or treatment suggest that misconceptions and gaps in knowledge about cancer and its treatment can have costs for AYA later in life. Several authors report associations between unmet information needs and psychological distress or poor Health-Related Quality of Life.[27-31]

Recent years have seen significant developments in AYA cancer care, and, in the United Kingdom (UK), AYA's access to specialist (AYA) cancer services has increased.[32] Nevertheless, scope remains to enhance AYA's preparation for, and support them through, the substantial challenges presented by cancer diagnosis and treatment. Improved understanding of AYA's prior knowledge (of cancer and treatment) might facilitate this, providing a foundation for the development of additional educational/informational interventions. Hence, in undertaking the analysis reported here, our objectives were to explore:

- AYA's views on their cancer knowledge prior to diagnosis, including its nature, extent, and source(s);
- AYA's views on whether and how this knowledge affected their experiences of diagnosis and care.

2 | METHODS

In reporting our methodology/methods, we take direction from the Consolidated Criteria for Reporting Qualitative Studies (COREQ).[33]

2.1 | Study design

Our work is underpinned by a critical realist perspective—a philosophical orientation combining a realist ontology and constructivist epistemology.[34] Qualitative description, focussed on the identification and description of minimally-theorized themes, provided a methodological framework for the study.[35,36] This involved an emergent, inductive design, purposive sampling, semi-structured interviews and an iterative relationship between data collection and analysis. Work was informed throughout by discussions with study advisory group members, who included AYA treated for cancer. The South East Scotland Research Ethics Committee 01 approved the study (REC reference 17/SS/0077).

2.2 | Study context

We undertook this study in Scotland, UK. Here, people diagnosed with cancer aged 16 or above typically receive treatment/care from oncologists and/or haematologists in a National Health Service (NHS) Scotland adult hospital. Some patients aged 16-19 receive care from paediatric oncologists in a paediatric hospital. Dedicated

AYA cancer/chemotherapy units, with specialist nurses and facilities, have been established (in adult and/or paediatric hospitals) in the four largest Scottish health boards. AYA living in other regions/health boards may be—and increasingly are—referred to these units/centres for treatment.

2.3 | Study sample

Our approach to sampling was purposive, seeking variation in characteristics potentially relevant to participants' experience(s) and perspective(s). These characteristics included age, gender, diagnosis and place of care. Our sample included AYA who were receiving/received front-line care in all the types of care setting described above. More information on the sample is provided in Table 1.

2.4 | Recruitment

Direct care team members made the initial approaches to AYA. These professionals outlined the study verbally and gave interested individuals a pack containing a participant information sheet, consent form and an opt-in form with a pre-paid envelope addressed to the research team. AYA interested in taking part were also given the option of contacting the project researcher (RH) by email or phone. RH followed up these expressions of interest, using contact details provided by AYA. This approach was intended to minimize pressure on AYA to participate, and the burden/work falling on direct care teams. However, it has meant we are unable to say how many potential participants were approached, or what their reasons for declining involvement were. Recruitment continued until our sampling ambitions were satisfied, and new topics and themes no longer emerged in new data.

2.5 | Data collection

Interviews were conducted between November 2017 and December 2018, at a time and place chosen by AYA. This was typically their home or a private space at their usual treatment centre. Whilst AYA were usually interviewed on their own, interruptions (by family and/or health professionals) were common. Four chose to be interviewed with a caregiver present. Interviews were conducted by RH, a social scientist with 15 years' experience of doing qualitative research, substantially relating to AYA's experiences of health and illness. RH was not known to interviewees prior to the study but outlined her professional background and research interests, as well as checking AYA's understanding of the project, before obtaining their written consent to participation. In the UK, people aged 16 years and above are entitled to consent to health research (and indeed to their own health care/treatment); hence, parental consent was not required. Interviews followed a topic guide, informed by literature reviews, and inputs from clinical co-investigators and AYA advisors. A copy of the full topic guide is appended to our previous publication.[14] Topics/questions of most relevance to this analysis were as follows: *Before your own diagnosis, what did you know about cancer and its treatment? Is there anything you wish you had known about cancer and its treatment? What have you learnt, since diagnosis, about cancer and its treatment?* The guide was revised over the course of the study to take account, and enable further exploration of, emerging themes. It was also adapted, in situ, in response to interviewees' accounts and to give AYA scope to talk freely about issues they viewed as important. Typically interviews lasted one to two hours; all were recorded, with participants' consent. Contemporaneous notes were taken, which included information on the environment/setting, interruptions and non-verbal communication.

TABLE 1 Participant characteristics

AYA with cancer (n = 18)	
Age: median (range)	
At diagnosis	19 (16-24) y
At interview	20 (17-26) y
Gender, male: n (%)	14 (78)
Ethnicity: n (%)	
White British	14 (78)
Non-white British	2 (11)
White non-British	2 (11)
Education/employment at diagnosis: n (%)	
Employment/work-based training	6 (33)
School/college	6 (33)
Undergraduate studies	3 (17)
Not in education/ employment	3 (17)
Diagnosis: n (%)[a]	
Bone sarcoma	6 (33)
Leukaemia	4 (22)
Germ cell tumour	3 (17)
Other sarcoma	2 (11)
Lymphoma	1 (6)
CNS tumour	1 (6)
Melanoma	1 (6)
Diagnostic type: n (%)	
Primary cancer	16 (89)
Relapsed cancer	2 (11)
Place of care: n (%)[a]	
AYA cancer unit in an adult hospital	14 (78)
AYA cancer unit in a paediatric hospital	3 (17)
Adult cancer service in an adult hospital with no AYA cancer unit	1 (6)
Reported enrolment in a trial: n (%)	5 (28)
Interviewed without caregiver present: n (%)	14 (78)
Time from diagnosis to interview: median (range)	10 (2-59) mo

Abbreviation: AYA, adolescents and young adults.
[a]Percentages do not sum to 100% due to rounding.

2.6 | Data processing and analysis

Interviews were transcribed verbatim, but anonymized (i.e. participant identifiers were removed). Transcripts were then imported into the qualitative data-indexing software NVivo (Version 11, QSR International Pty Ltd., Doncaster, Victoria, Australia). After familiarization with the transcripts (involving close reading and line-by-line coding), more focussed coding and analysis was undertaken by two members of the qualitative research team (RH and JL). Using a constant comparative approach,[37] these individuals coded for topics, issues and themes. Mapping and memo-ing strategies were used to define the content, parameters and relationships between codes. RH and JL prepared analytical reports, with these providing a basis for discussions to refine and agree coding and reporting frameworks. Whilst some topics were of a priori interest, and informed lines of questioning in all interviews, the themes/sub-themes reported in this paper were largely emergent, that is derived from the data. These are outlined in Figure 1 and detailed in the Results section. Members of the wider research team confirmed that the identified themes were reflective of the data. Whilst interviewees were not asked to comment on individual transcripts, they, and other AYA diagnosed with cancer, were invited to provide feedback on key study findings via a workshop organized in the final stages of the project.

3 | RESULTS

3.1 | Study participants

We interviewed 18 AYA diagnosed with cancer whilst aged 16-24 years and receiving care (active treatment and/or follow-up) through an NHS Scotland oncology/haematology service. 17/18 were being (or had been) treated in a specialist AYA cancer/chemotherapy unit. Further information is provided in Table 1.

3.2 | Study themes

Under the over-arching theme of '(Views on) knowledge of cancer and its treatment prior to diagnosis', we identified a series of contributory themes. These themes and their relationships are mapped out in Figure 1: those highlighted/shaded are then detailed in narrative form, below. We begin by documenting (Theme 1) how AYA characterized their knowledge of cancer and treatment ahead of their own diagnosis and care. We note where AYA identified important differences between their prior understanding and subsequent realities and consider their explanations for perceived gaps in knowledge. We then document (Theme 2) the consequences AYA perceived these knowledge gaps as having had: pre-diagnosis (sub-themes 2.1 and 2.2); at diagnosis (sub-theme 2.3) and post-diagnosis/on treatment initiation (sub-theme 2.4). Theme headings are reflective of the content of participants' accounts: verbatim quotes are indicated by inverted commas.

3.2.1 | Theme 1: Characterizing prior knowledge as limited and superficial

AYA consistently characterized their prior knowledge of cancer and its treatment as limited, superficial and/or abstract. Even those who portrayed themselves as having had 'some' previous knowledge typically drew attention to its shortcomings, using words such as 'basic', as illustrated by A04, a young man diagnosed shortly after starting university:

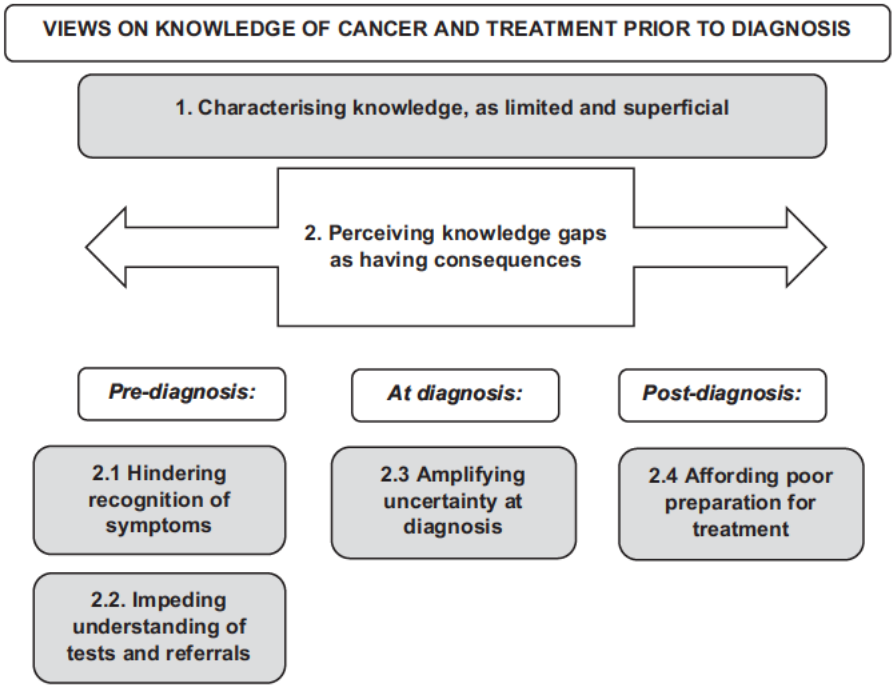

FIGURE 1 Emergent themes

> I knew the basics... I knew it was like, a serious disease, that affects your body's cells, like and the way that they divide and stuff.
>
> (A04)

Others described how it had only become clear to them retrospectively quite how deficient their knowledge had been. For example, one young man, interviewed towards the end of treatment, commented:

> It's amazing like... if you think you know anything about cancer, or that, when you actually get it... what you think you know, is, like, a fraction of what is actually there... you just sort of see on the surface, for what it is, like this really negative, horrible thing.
>
> (A16)

Reflecting on their knowledge of cancer treatment, interviewees noted that this had been skewed towards chemotherapy and even here had been limited. For example, they had not appreciated the variety of regimens in use:

> If you're not familiar with like cancers, then you just think chemotherapy's one thing. And that there's not like, hundreds of different types of chemotherapy... and that you could get them in tablet form and things like that.
>
> (A06)

Adolescents and young adults also reported limited prior knowledge of how chemotherapy (and other cancer treatments) worked, and what the potential long-term implications of such treatments might be. Several, for instance, commented that they had been entirely unaware of the potential impact on their fertility:

> (Going) back to like my, my lack of understanding about cancer and stuff, I had no idea fertility was an issue.
>
> (A15)

Discussion of the sources of AYA's knowledge provided some explanation for its character and limitations. For example, interviewees recalled receiving some education about cancer and its treatment at school, which they now viewed as extremely rudimentary:

> At primary school I got a book which was (called)... the "Big Book of Science" or something... and it had this, just this little bit about chemotherapy, which I found fascinating, and... this little like... pictogram of the coloured, drugs, finding the cells, and killing them...
>
> (A14)

In addition, AYA described learning about how cancer manifests, is experienced, and treated, via the media. Reflecting on how media portrayals had shaped their understanding and expectations of cancer treatment, AYA noted a focus on chemotherapy, and recalled TV and film depictions of people 'hooked up' to a machine, like 'the guy in "Breaking Bad"' (A14). In retrospect, AYA judged such portrayals as misleading:

> You see stuff on the telly, like, and you think to yourself, "No, that's, that's not at all what it's like!"
>
> (A03)

> TV (shows) cancer patients in bed, with a towel around their head... It's not what the TV, the films, make it out to be.
>
> (A11)

Typically, AYA talked of having had very little direct contact with other people with cancer prior to their own diagnosis. Some identified family members or family friends who had (had) cancer, but these were usually older, and often distant, relations with whom they had had little or no contact over the course of treatment:

> (Girlfriend), her Gran unfortunately passed away with cancer a couple of years ago... that's the only time I've really experienced cancer.
>
> (A16)

Even when AYA reported knowing (of) another AYA with cancer, they seldom portrayed themselves as having accumulated and drawn on knowledge about that person's experience. Accounting for this—and their lack of knowledge more generally—interviewees explained that, until their own diagnosis, cancer had seemed a topic of little personal relevance:

> You just never think it's gonna happen to you. Like you just think it's... one of these things, like, "Oh it's such a shame that (so-and-so) got diagnosed, but it's, no, it'll never happen to me!"
>
> (A05)

3.2.2 | Theme 2: Perceiving knowledge gaps as having consequences

3.3 | Sub-theme 2.1: Hindering recognition of (signs or) symptoms

Interviewees suggested that their limited knowledge had had consequences at different points in their cancer journey. These included not recognizing emerging signs/symptoms as potentially indicative of cancer. In particular, many interviewees reported not having

appreciated the significance of non-specific symptoms such as fatigue, which some described discounting for long periods:

> Before like getting ill and that, I'd come home from school, and go for like a five-hour nap. And... Mum'd be like, "Why are you going to bed?" And I'm like, "'Cause I'm tired". You never really think about things... you kind of just brush it off... you don't think (it might be cancer).
>
> (A18)

Others noted how they had remarked and reflected upon such symptoms, but—initially at least—attributed them to life pressures and/or lifestyle. For example, a young man diagnosed shortly after graduation described how he had ascribed fatigue to the demands of his examinations. Similarly, a young man who had been working in a bar when his health had started to deteriorate explained how he and a friend had attributed his cough to a hedonistic lifestyle:

> I had this cough for quite some time... I told my friend... and she told me, "Just drink less"... (That) helped, so I thought, "Aah, it's not that bad. (Laughs) It can't be cancer, right?"
>
> (A13)

Adolescents and young adults described how these mundane explanations had encouraged stoicism, self-management (eg rest, reduction in alcohol consumption) and monitoring. Typically, they reported seeking help from health professionals (eg visiting a GP) only once self-care strategies had proved ineffective, symptoms became more pronounced and/or a caregiver intervened. By this point, some AYA said they had been very unwell. Even so, some noted how they had only given partial (selective) reports, prioritizing their most troubling and/or specific symptoms:

> So I went to this doctor's appointment. Talked to the doctor, told her my symptoms. Now I wasn't, I didn't mention some of the things I've mentioned to you, because, I thought maybe they weren't that important, so, I think it would've definitely been hard for the doctor I saw to make, a complete diagnosis, just on my presenting complaints. However, she probably could've done a better job at fishing the information out of me, and maybe asking more questions.
>
> (A12)

3.4 | Sub-theme 2.2: Impeding understanding of the significance of referrals and tests

Adolescents and young adults suggested that their lack of knowledge had had consequences even after help from professionals had been sought. Whilst they often described referral to secondary care for specialist attention and/or further investigations as prompting a suspicion that something quite serious was wrong, few reported having considered cancer as a possibility until this was explicitly suggested by a health professional. Hence, even when scans/imaging, or more invasive investigations such as biopsies had been scheduled, this had not necessarily set off 'alarm bells', as the following young man, who had been experiencing pain and swelling in a lower limb described:

> It's strange though, even though I had a biopsy, the idea that I might have cancer... barely crossed my mind.
>
> (A15)

Furthermore, AYA noted how they had not necessarily understood the results of investigations as being suggestive of cancer. Some remarked that the language professionals used to report the discovery of abnormalities had been unfamiliar to them. For example, A17, a young man 'blue-lighted' from his local hospital to a larger regional facility for investigation of a suspected neurological cancer, explained that the term 'mass' had not held a clear meaning for him:

> They said, "There's a mass in your brain," or something. [Interviewer: And what did you take that to mean?] No idea... "A mass" – that could mean anything.
>
> (A17)

Adolescents and young adults viewed their limited appreciation of the significance of referrals, tests and results, as having important practical and psychological sequelae. For example, some reported that misunderstanding of the reasons for referrals and/or tests had led to them attending pivotal consultations alone, including those at which their cancer diagnosis was first disclosed:

> I went by myself (to get the results of biopsy), because, like I say, I wasn't expecting... [Interviewer: That sort of news?] Yeah, I was, I thought it would be relatively benign.
>
> (A15)

Other AYA, ultimately diagnosed with haematological malignancies, described their bewilderment and/or distress on seeing 'Cancer' signs when admitted to hospital:

> I was a bit confused. I didn't really know what was up with me. And then obviously at the (Cancer Centre), it says (Regional) Cancer Centre, whatever it is, underneath the (Hospital Name). So I was like, "Right, what's, what's going on?".
>
> (A07)

3.5 | Sub-theme 2.3: Amplifying uncertainty at diagnosis

Adolescents and young adults explained how lack of awareness that cancer was a plausible explanation for their symptoms had left them both practically and emotionally ill-prepared for diagnosis. Many suggested that the confusion, shock and distress of diagnosis had been compounded by their lack of knowledge of the types of cancer involved. Few AYA reported having had any familiarity with their/common AYA cancers (sarcomas, leukaemias, brain tumours, testicular and skin cancers). Some noted how, on receiving their diagnosis, they had not realized they were being told they had cancer:

> When (Consultant Haematologist) told me first, It's, it's Hodgkin's Lymphoma, he put it, the name through Google Translator – and I still didn't know what it, what it was.
>
> (A13)

> The word sarcoma isn't part of my vernacular so much that I instinctively identified it with cancer, so I needed a bit… of explaining, and then (the surgeon) said, "Your cancer is…" and I'm like, "Oh crap, right, so this is cancer?"
>
> (A15)

With a few exceptions (typically older interviewees), AYA portrayed themselves as having been profoundly unsure of the implications of their diagnosis, and the sort of future they might expect. Reflecting on the moment she learnt she had cancer, A18 explained:

> (Mum) told me. She was like, "You've got cancer". And I just… kind of sat there… It's just a… a word that you don't really know how to be… 'cause, you know, you don't really know the outcome to that.
>
> (A18)

Nevertheless, AYA reported having had to rapidly make important decisions about their care and the practical organization of their lives.

3.6 | Sub-theme 2.4: Affording poor preparation for the realities of treatment

As reported in Theme 1, AYA perceived their prior knowledge of cancer treatment as having been minimal. AYA suggested that this too had had consequences, highlighting the gulf between their expectations of treatment and actual experience. For many, treatment had involved not only chemotherapy, but also radiotherapy and/or surgery. Several commented that they had been unaware of the duration of regimens (commonly months and for some AYA-relevant cancers as much as three years). This, and/or the need for frequent and numerous visits including (for some) extended in-patient stays, had been unexpected and dismaying:

> The biggest shock… was the length of time that I was gonna have chemo… the fact that it was gonna go on for months and months was a bit of a surprise.
>
> (A08)

Though AYA acknowledged health professionals' efforts to make them aware of the treatment plan and potential side effects, many said they had not grasped the pervasive and brutal impact treatment would have on their lives. With a few exceptions, AYA described treatment as far more debilitating and disruptive than they had anticipated. Several reflected on how, at diagnosis, they had viewed cancer as a 'blip' and treatment as a temporary disruption. Some described planning things to do during treatment, and, as A15 explained, 'almost treating this like a sabbatical'. These AYA said they had soon realized that few of their ambitions were achievable:

> As a symbol of how naïve I was, when I first got diagnosed… I was thinking about how I could do push-ups, and little workouts in my room, to keep my body fit, and, I did that for like a week, and then I couldn't do it anymore. Because I had this vision of having my body intact, over this period, but, there was just, there was no chance of that happening.
>
> (A12)

In contrast to their initial expectations, AYA reported how their treatment regimens had proved all-consuming. Some explained how the effects of treatment had presented challenges to the most fundamental aspects of daily life, such as taking a shower:

> You don't realise the implications… all the ways that it affects you… like some of them are… they're so, I wouldn't say basic, but they're things that maybe we take for granted, on a daily basis kind of thing.
>
> (A16)

To better prepare AYA for the realities of treatment, some interviewees suggested that health professionals should have much franker conversations at the time a plan was discussed and agreed. However, others expressed different—and sometimes conflicting—attitudes to such information. These AYA surmised that knowing what lay ahead might have compounded their anxieties.

4 | DISCUSSION

Our objectives in this analysis were to explore AYA's views on their cancer knowledge prior to diagnosis, including if, and how, they felt that this knowledge had affected their experiences of

diagnosis and care. In so doing, it was not our intention to suggest that knowledge is the only factor affecting experience, but simply a factor which, to date, has not perhaps received the attention it deserves. Our analysis revealed that AYA consistently characterized their knowledge as limited and superficial prior to diagnosis. We have further highlighted how AYA perceived inadequate knowledge as having had consequences prior to, at, and following diagnosis: hindering recognition of symptoms; impeding understanding of tests and referrals; amplifying uncertainty at diagnosis; and affording poor preparation for the harsh realities of treatment.

Whilst our research design does not equip us to confirm causal relationships, these findings have sensitized us to the potentially serious implications of (lack of) knowledge and prompted consideration of the possibilities for intervention. Three findings in particular warrant discussion: lack of knowledge may hinder symptom recognition, delaying help-seeking and diagnosis (see sub-theme 2.1); lack of knowledge may amplify the shock, uncertainty, and distress of the diagnostic process (see sub-themes 2.2 and 2.3); and lack of knowledge may compound the already significant difficulties of the treatment experience (see sub-theme 2.4).

Our observation that lack of knowledge may hinder symptom recognition is consistent with findings from both qualitative work with older but still relatively young adults (24-35 years) diagnosed with cancer [9] and from survey-based research with healthy AYA.[15-18] However, our work advances that literature by illuminating reasons for AYA's limited knowledge, and highlighting potentially serious consequences, including delays in help-seeking, and incomplete reporting of symptoms on presentation. The latter finding underscores the importance of professionals in primary and/or emergency care taking a complete and detailed history, and encouraging AYA to disclose any/all symptoms.

Less well documented previously is how AYA's lack of clarity regarding the reasons for referrals and tests can have important and undesirable consequences, such as AYA attending pivotal appointments alone.[9] Again, our work extends understanding of how such situations may arise. For example, our data suggest that professionals' use of ambiguous and/or unfamiliar language may compound lack of knowledge and foster misunderstanding. Whilst professionals may, understandably, seek to avoid distressing AYA before a diagnosis is confirmed, lack of awareness of the diagnostic trajectory may also have serious consequences. Ensuring AYA (and indeed other patients) understand fully what is happening to them and why would seem fundamental to quality care.[38,39] Hence, where cancer is suspected, professionals might consider using less ambiguous language, checking understanding and advising AYA to bring a family member or friend to subsequent consultations.

Our finding that lack of knowledge may contribute to the difficulty of AYA's treatment experience is quite new. Prior research suggests that AYA are not alone in lacking knowledge of contemporary cancer treatments. These are complex and varied, and similar assertions have been made about knowledge in other patient/research populations.[40-42] However, the impact of lack of knowledge cannot be assumed to be the same for all patient/age groups. We believe this topic invites further exploration.

4.1 | Implications for policy and practice

Our findings prompt important questions about whether, when and how, AYA's knowledge of cancer might be enhanced, with a view to promoting timely diagnosis and optimizing experiences of treatment/care. Two broad approaches warrant consideration: universal education on AYA cancer, and/or health; and targeted education (enhanced information and counselling) at and after diagnosis.

Other researchers have previously argued for, the introduction and/or expansion of developmentally appropriate, school-based education on cancer risks and symptoms.[16] Evaluation of the effectiveness of such initiatives for increasing (younger) adolescents' knowledge of cancer risk factors and symptoms has found evidence of some short-term success.[43] However, our findings suggest some barriers to lasting impact might be anticipated. Firstly, as our interviewees reported viewing cancer as a topic of limited personal relevance prior to diagnosis, the effectiveness of such educational initiatives may be contingent on changing this mindset. Moreover, the non-specific nature of many of the symptoms reported by AYA (eg fatigue) may make consistent and enduring recognition hard to achieve. Other work, on delays in diagnosis of haematological malignancies in adults, has reported similar challenges and recommended that education should focus on promoting recognition of normal health (encouraging help-seeking should that change).[44] Such an approach might have salience for AYA cancers too. Furthermore, this sort of message might usefully be targeted at both AYA and the key adults in their lives (e.g. parents, caregivers, teachers, employers).

On diagnosis with cancer, our findings suggest that (for most AYA) the learning curve will be steep, requiring assimilation of both new language and concepts. AYA's reflections on their experiences of diagnosis and initial/front-line treatment point to the importance of education at and beyond diagnosis, but also to the challenges of delivering this. We have discussed elsewhere how AYA's physical and emotional states can create difficulties processing and absorbing information when first diagnosed.[14] Whilst there are clearly significant obstacles to enhancing knowledge at this time, there may be more scope to achieve this over subsequent weeks and/or months, once AYA have processed their diagnosis and acquired some experiential understanding of treatment. Most of the AYA who took part in our study were receiving care through specialist AYA cancer services and supported by committed professionals with some level of expertise in providing care to this (age) group. Nevertheless, their accounts suggest their educational/informational needs were not fully met. Other authors have reported that (adult) oncologists do not consistently ascertain their patients' prior knowledge and argued

that such a practice is essential if information is to be tailored to patients' evolving needs.[45] The AYA population too might benefit from clinicians establishing a practice of routinely assessing individual patients' prior—and evolving—knowledge and informational needs. Consideration might also be given to the role that parents/caregivers can play in building AYA's cancer knowledge, although it is important to note that parents/caregivers' priorities and information needs may not always align with those of AYA.[14]

4.2 | Strengths and limitations

The retrospective nature of our study might be considered both a strength and a limitation. Unlike previous survey research with healthy AYA, we collected data from AYA with cancer, who, in contrast to their (healthy) peers were in a position to identify 'known unknowns' (ie what, of importance, they had not previously known). In addition, the use of semi-structured interviews allowed AYA the flexibility to raise, and describe, issues viewed by them as particularly salient—indeed it is as a result of this that 'prior knowledge' emerged as a significant analytical theme. However, recall difficulties may have limited the range of comments/responses, and constrained our understanding of how AYA's perceptions of their knowledge, and need for this, may change over time. Prospective, longitudinal work, involving serial interviews, might provide additional and useful insights on 'teachable moments', that is when AYA with cancer are particularly desirous of, and receptive to, new information and knowledge.

5 | CONCLUSION

This qualitative work illuminates how (lack of) prior knowledge of cancer may impact upon AYA's experiences of diagnosis with cancer and initial/front-line care. It suggests knowledge gaps may have serious consequences at various points in AYA's pathways to and through treatment. These findings prompt questions as to whether, when and how, AYA's knowledge of cancer and its treatment might be improved. Unfortunately, there are no easy answers, though we suggest two broad approaches warrant consideration. Ultimately individuals may vary in their keenness for, and receptivity to, cancer-related information at different times[38]; assessment and repetition may be key to improving AYA's knowledge of cancer and its treatment/care.

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CONFLICT OF INTEREST

We have no conflicts of interest to declare.

DATA AVAILABILITY STATEMENT

Data (eg transcripts) have not been made not publicly available, due to the small number of AYA diagnosed with cancer in Scotland each year, and the associated risk of participants being identified notwithstanding the removal/redaction of obvious identifiers. Requests, to the corresponding author, for access to the data underpinning this paper will be considered and accommodated where reasonable.

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ORIGINAL ARTICLE

Strategies for improving access to clinical trials by teenagers and young adults with cancer: A qualitative study of health professionals' views

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ABSTRACT

Objective: Few teenagers and young adults (TYA) with cancer participate in clinical trials. Lack of opportunity has been identified as a major barrier. We canvassed health professionals' views on how TYA's access to trials might be improved.

Methods: We interviewed 35 professionals with responsibility for delivering or facilitating cancer care and/or clinical trials. We analysed data using a qualitative descriptive approach.

Results: Interviewees viewed improving TYA's access to trials as challenging, but possible. They reframed the problem as one of rare disease and surmised that modifying the organisation, administration and resourcing of research (and care) might expand opportunities for both TYA and other patients with low volume conditions. Proposals coalesced around four themes: consolidating the pool of patients; streamlining bureaucratic requirements; investing in the research workforce; and promoting pragmatism in trial design.

Conclusion: Accounts suggest there is scope to improve access to trials by TYA with cancer and other patients with rare diseases. Though re-configuring care, research and resource frameworks would present substantial challenges, doing nothing would also have costs. Change will require the support of a range of stakeholders, and agreement as to the best way forward. Further work, such as priority setting exercises, may be necessary to reach a consensus.

KEYWORDS
cancer, clinical trials, qualitative research, rare diseases, teenagers, young adults

1 | INTRODUCTION

Clinical trials have played a decisive role in advancing cancer treatments (Keating & Cambrosio, 2007). However, teenagers and young adults (TYA, aged around 16–24 years) have had low levels of trial involvement; this is thought to have hindered progress in improving survival (and other) outcomes in this patient group (Bleyer et al., 2005; Bleyer et al., 2007). A growing body of work

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has sought to explain TYA's limited trial involvement, with a diversity of barriers identified (Barakat et al., 2014; Bell et al., 2018; Fern et al., 2014; Forcina et al., 2018; Freyer & Seibel, 2015; Friend et al., 2016; Pearce et al., 2018). Lack of opportunity has been identified as an important—albeit not exclusive—obstacle to participation (Burke et al., 2007; Fern et al., 2014; Freyer & Seibel, 2015; Tai et al., 2014).

The nature of the cancers with which TYA typically present is an important factor in the lack of relevant trials and their limited availability at the places where TYA receive cancer care. The malignancies most typically seen in TYA (soft tissue sarcomas, bone cancers, acute forms of leukaemia, cancers of the lymphatic system and brain tumours) are nonetheless rare: securing funding for rare disease trials presents considerable challenges. Moreover, where the trial-eligible population is small and recruitment therefore predicted to be (s)low, sponsors may seek to open trials only at the very largest centres. Local decision-making may further affect opportunities, with clinicians prioritising engagement with trials in higher volume diseases (Hart et al., 2020). Hence, even when relevant trials are established, they may be opened only at a limited number of centres and TYA's access may depend substantially upon where they receive care (Fern et al., 2014; Fern & Taylor, 2018; Hart et al., 2020).

Whilst barriers to TYA's enrolment in cancer trials are relatively well-documented, a recent systematic review highlighted a paucity of studies attending to *facilitators* (Siembida et al., 2020). Though various commentators have hypothesised how access might be improved (Burke et al., 2007; Davis et al., 2017; Freyer & Seibel, 2015; Gupta & Indelicato, 2014), empirically informed recommendations remain limited. Further investigations are urgently needed, as, where relevant cancer trials have been established and widely opened, marked improvements in participation have been achieved (Fern et al., 2014; Fern & Taylor, 2018). Hence, there have been calls for researchers to do more to improve understanding of, and provide practical guidance on, the specifics of interventions which might promote participation (Fern & Taylor, 2018; Friend et al., 2016). Identifying effective strategies to improve TYA's access to clinical trials was recently agreed, via a stakeholder partnership priority setting exercise, to be one of the top three priorities for TYA cancer research in the UK and beyond (Aldiss et al., 2018). The importance of such work has been further highlighted in an editorial in this journal (Stark et al., 2018). In line with such calls, we canvassed professionals' views on practical strategies which might improve TYA's access to cancer trials. Whilst our study was conducted in Scotland, UK, we sought to generate recommendations which might have relevance to similar (small and dispersed) patient populations elsewhere.

2 | METHODS

Our qualitative interview study featured an emergent, inductive design, purposive sampling and an iterative approach to data collection and analysis (wherein findings from early interviews were used to inform subsequent sampling decisions and areas explored in later interviews). Such an approach allows for, and indeed encourages, the uncovering of novel and unanticipated findings. The study design was approved by the South East Scotland Research Ethics Committee 01 (REC reference 17/SS/0077).

2.1 | Orientation

Our methodological orientation was qualitative description, a pragmatic approach well-suited to applied research, and focused on the identification and description of patterns and explanatory themes (Sandelowski, 2000; 2010).

2.2 | Setting/context

The study took place in Scotland, UK. National Health Service (NHS) Scotland operates through 14 regional health boards (HB), 13 of which agreed to host/support the study. Cancer care is delivered at more than 20 hospitals, though certain services/treatments, including specialist TYA units, are only available at major hospitals within the four largest health boards. Around 180 TYA are diagnosed with cancer in Scotland each year. Most receive treatment in adult hospitals, but paediatric hospitals (three) deliver care to some 16- to 19-year-olds. NHS Research Scotland (NRS) provides some infrastructure for research.

2.3 | Sampling and recruitment

We sampled professionals purposively, seeking variation in: roles; sub-specialties; and the services, hospitals and health boards within which interviewees worked (for example, small, large, urban, rural, with and without specialist TYA services/facilities). Early findings prompted us to increase both the size and the diversity of the pool of interviewees. In particular, we extended our capacity to thoroughly explore structural factors, by including a small number of professionals with research design, oversight or support responsibilities. Potential interviewees were suggested by: clinical members of the research team; advisory group members and other colleagues; and interviewees. They were approached, initially, in writing, by either the chief investigator (AJ) or the project researcher (RH), with all follow-ups made by the latter. Of the approximately 70 professionals approached, around half opted in. Reasons for non-participation, where given, generally related to the pressures of work and/or infrequent contact with TYA patients. We discontinued recruitment once the core team (AJ, JL and RH) were in agreement that sampling ambitions had been satisfied and data saturation had been reached (where no new issues or themes were identified in additional data collected).

2.4 | Data collection

Data were collected using semi-structured interviews, a format enabling participants to raise issues they considered salient, including those unforeseen at the study's outset. Interviews followed a loosely structured topic guide (Appendix S1), the development of which drew on the literature, the team's expertise and insights from advisory group members. This topic guide was revised during data collection to take account of early findings. Questions were also tailored in situ to accommodate interviewees' varying roles, experiences and perspectives. Hence, the guide should be considered *indicative* of the topics explored, not a script. All interviewees were given written information on the study and provided written consent. Interviews were conducted by RH, a social scientist with more than 15 years' experience of conducting qualitative research, substantially relating to health and health care. RH was not known to interviewees prior to this study. Dependent on interviewees' preferences, interviews were undertaken face-to-face (in a private space) or by telephone. Most lasted 45–60 minutes. All took place between December 2017 and August 2018.

TABLE 1 Participant characteristics

Professionals interviewed		**35**
If had direct care responsibilities	Yes	30
	No	5
Type of healthcare professional	Consultant	23
	Nursing	4
	Other	3
Consultants' specialisms	Clinical oncology	7
	Medical oncology	6
	Haematology	6
	Paediatric medicine	3
	Surgery	1
If had research oversight or support responsibilities	No	25
	Yes	**10**
Type of responsibilities	Research governance/ facilitation	5
	Senior role in a CTU or CRF	3
	Specialised/technical trial support	2

2.5 | Data processing and analysis

All interviews were audio-recorded and transcribed verbatim. Our approach to analysis was qualitative description, directed towards the production of detailed, lightly theorised accounts of participants' experiences and perspectives (Sandelowski, 2000; 2010). We began with an initial period of data immersion, after which preliminary 'line-by-line' coding was undertaken by two members of the team (RH and JL). RH and JL then agreed on a provisional framework for more focused coding. Specialist software (NVivo (version) 11 (QSR International Pty Ltd., Doncaster, Victoria, Australia)) was used to facilitate this task. Following several rounds of progressively more focused coding, informed by the principle of 'constant comparison' (Strauss & Corbin, 1990), RH and JL each prepared an analytical report; these were compared and discussed. Points of difference (which were limited) were reconciled through discussion and additional scrutiny of the data. This (additional scrutiny) involved RH systematically reviewing and coding all data to confirm and clarify the limits of patterns, subtle variations and deviations from these. Diagrams, charts and short reports were used to give the wider research team and advisory group members (who included research participants and other professionals) access to key findings and supporting data. Their feedback was used to refine our interpretation of the findings.

3 | RESULTS

We interviewed 35 professionals working in a variety of roles, subspecialisms and settings. 30/35 had some clinical/direct care responsibilities whilst 10/35 had research design, oversight or support responsibilities (e.g., a senior role in a clinical trials unit or research facility, research governance/facilitation responsibilities and/or provided specialised/technical trial support). Interviewees were drawn principally from six Scottish health boards. However, several provided services in other areas or had previously worked elsewhere. Further details can be found in Table 1. Unique identifiers, used in reporting, below, are prefaced as follows: DC-C/N/O- (Direct Care-Consultant/Nursing/Other); ROS- (Research Oversight or Support only).

In short, interviewees portrayed extending TYA's access to cancer trials as challenging, but surmised there was potential to achieve that. They (re-)framed the problem as an issue of rare disease, rather than of age *per se*, reflecting the wider challenges of delivering trials to small and dispersed populations. Hence, they suggested, strategies modifying the organisation, administration and resourcing of research (and care)—if workable and effective—might expand access to trials for both TYA *and* other patients with low volume conditions. Interviewees made a range of proposals which coalesced around four inter-related themes: consolidating the pool of patients; streamlining bureaucratic requirements; investing in the research workforce and promoting pragmatism in trial design (Figure 1). We begin by considering how interviewees reframed the problem, before outlining their potential solutions.

3.1 | Reframing the access issue: a problem of rare disease ('it's about numbers')

How interviewees conceptualised the 'problem' provided a logic for the solutions they proposed. Early in interviews, many questioned whether framing the issue of limited access to cancer trials as a TYA/

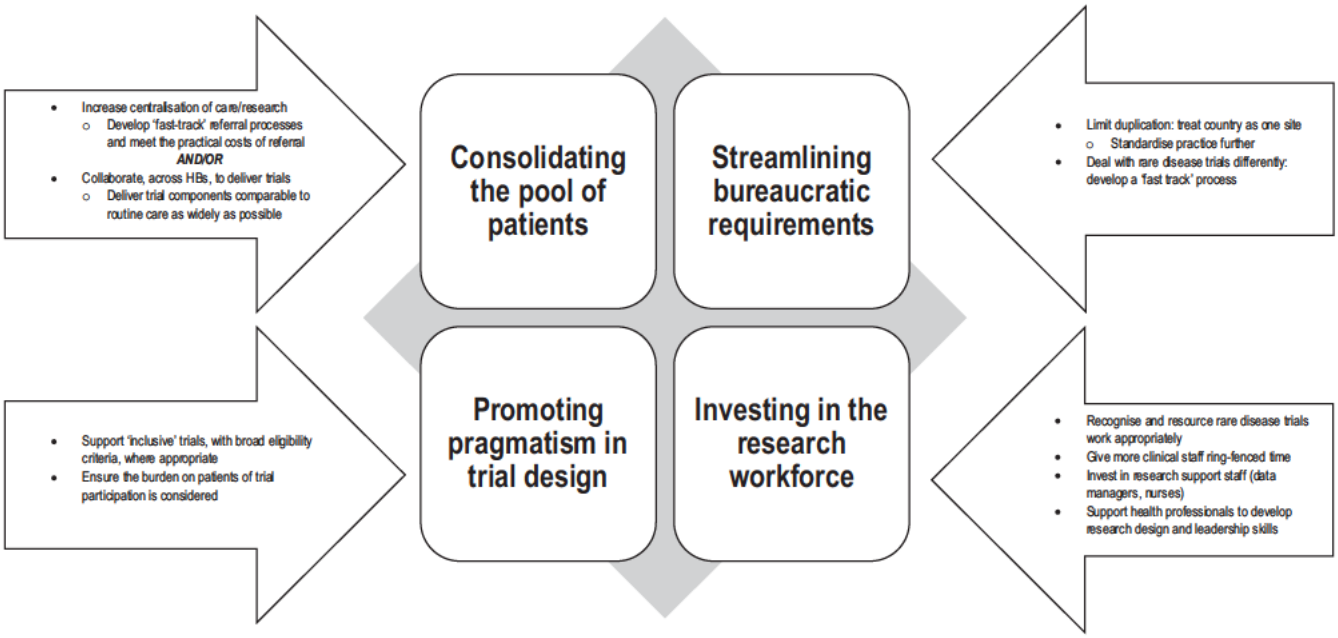

FIGURE 1 Strategies for improving TYA's access to cancer trials.

age-related matter was appropriate or helpful. These professionals contended that the problem was one of rare disease, and numbers, not patient age *per se*. They considered other, older patients with rare cancers to have similarly limited access to trials.

Interviewees explained that, where patients were few and widely dispersed, the (already significant) costs of delivering trials were amplified, by, amongst other things, the need to negotiate and support the involvement of a wide cast of professionals and organisations. They viewed the returns on such investments as highly uncertain, since relevant patients might ultimately never be seen by a proportion of those professionals or organisations. They considered the disincentives to establishing and hosting, these sorts of trials to be substantial. However, alongside noting the difficulties of delivering trials relevant to small and dispersed populations, interviewees highlighted the increasing importance of being able to do so, as medicine became more 'personalised' and targeted in its approach.

Interviewees explained how, in oncology, 'rare' disease was becoming a more pervasive concern, as 'common' malignancies were more precisely characterised, and patients divided into smaller sub-groups for treatment and care. In effect, through this process, 'common' cancers were being rendered 'rare'. Some interviewees stressed that this issue was not particular to the specialism, and treatment and trials generally were 'becoming more niche… That obviously affects everybody, not just TYA (with cancer)' (ROS-05). These interviewees stressed that if the use and development of targeted treatments were to continue, it was critical to find ways of making it viable to open trials at multiple sites, with modest and/or uncertain recruitment prospects:

> 'We're going to need to do more, small trials, because of just the nature of the new innovations that are coming through, (which are) tending to be more targeted to smaller populations. So we'll see a lot more trials that are recruiting smaller patient numbers … and therefore need more sites that are going to be prepared to open for one or two patients.' *(DC-C-26)*.

3.2 | Consolidating the pool of eligible patients

Interviewees identified a number of ways organisational features of care and research could impede or facilitate set-up and efficient recruitment into TYA-relevant (rare disease) trials, and speculated that 'if we think about it… we could organise ourselves better' (DC-C-25). Proposed strategies were orientated towards consolidating the pool of trial-eligible patients, via different forms of *centralisation* and/or *collaboration*.

3.2.1 | Centralising care and/or research

Some argued that to achieve the 'critical mass' of eligible patients needed for TYA-relevant (rare disease) trials to be viable, greater centralisation of care was needed:

> 'The difficulty with this age (group), it's small numbers, so setting up trials where, you know, you've got small numbers of patients, because of the expense, the only way round that issue is to centralise care.' (DC-C-13).

Others proposed centralising trials work, rather than routine cancer care, and questioned the wisdom of opening TYA-relevant trials at more than one specialist TYA unit:

> 'Another issue that I've had, is… somebody's come to us with a really rare tumour study, one patient

> every two years, a sarcoma study, say, and I've said, "Well, we'll do it, but as long as we're the only centre in Scotland, because we need to make sure that if there's a patient with the disease in Scotland, they come here..." What we can't do is open four centres in Scotland to capture this.' (DC-C-28).

Interviewees acknowledged that the success of either approach would depend on having processes in place to 'fast-track' referral, so treatment was not delayed. Whilst highlighting the potential benefits of centralisation, many also drew attention to its costs. Some interviewees questioned TYA patients' willingness and/or ability to travel, noting that trials varied in the provision made for patients' (and family members') expenses whilst securing financial support from health boards could be onerous and time-consuming. Additionally, interviewees often appeared conflicted about advocating care remote from patients' homes, highlighting the benefits of proximity to family and other social support.

3.2.2 | Collaborating in trial delivery

Some interviewees suggested that collaboration (between health boards or nationally) might afford similar benefits to centralisation, whilst limiting the costs and burdens to TYA and their families. Interviewees hypothesised that models of collaboration could take a variety of forms, with some proposing a 'hub and spoke' arrangement where a trial was opened through one centre, with those parts of the protocol comparable to routine care being delivered at 'satellite' sites under the first centre's oversight.

Whilst some interviewees anticipated reluctance on the part of sponsors and/or principal investigators to support such a model, a few countered that there were precedents and therefore this:

> 'might be something that we could actually explore for more trials – if there is a specific treatment or technique for a trial that we couldn't ever have up here, just, through logistics... would it be possible to send the patient (elsewhere) for that technique, but they could come back and have most of their treatment actually here? This might be something we can take forward.' (ROS-5).

Others drew attention to existing facilitators, such as histories of professionals working across health board boundaries to provide leadership or support for trials and/or delivering routine care as part of a regional service.

3.3 | Streamlining bureaucratic requirements

Interviewees talked at length of the time and other costs of regulatory and administrative activities, and how these might be reduced. Though the importance of strong governance procedures was recognised, there was a perception of contemporary approval processes as cumbersome, bureaucratic and a disincentive to opening rare disease trials widely. Interviewees stressed the importance of doing everything possible to ***limit duplication*** of tasks and standardise practices and suggested there might be ways of ***dealing with rare disease trials differently.***

3.3.1 | Limiting duplication of activity

Though interviewees with Research and Development (R&D) roles/responsibilities emphasised that efforts to streamline approval processes had already been made, their clinical/investigator colleagues expressed the view that more could, and should, be done to reduce duplication of work across health boards, favouring increased unification of activity where possible. Some referred to a concept of 'one-site Scotland', where, from a regulatory and administrative perspective, all (14) Scottish health boards might be treated as a single site/trial centre. It was acknowledged that the current structure of NHS Scotland presented challenges to this sort of arrangement:

> 'Each health board is an independent legal entity... I did put out the idea... (that) we should invent a new legal entity which was the whole of Scotland, which was specifically there to run trials. But clearly... there'd be lots of barriers to that as well. (DC-C-28).

However, interviewees suggested that, if such legal issues could be resolved, the burden on local investigators would be reduced, and the attractiveness of smaller centres to (commercial) trial sponsors increased. More modest benefits might be gleaned from further standardisation of policy, practice and contracting across health boards:

> 'We find ourselves always negotiating... with individual sites... the contract... is pored over the... the legal team of that particular site... they'll (all) come up with something they want changed, and it's often trivial, but it takes time... I would just love it, if there could be a more national system... a standard contract.' *(DC-C-26).*

3.3.2 | Dealing with rare disease trials differently

Interviewees stressed the disproportionate regulatory burden on rare disease trials, where recruitment was uncertain but always likely to be modest. Signing up to every new rare disease trial in case an eligible patient presented appeared neither feasible nor sensible. Instead, it was suggested that the rational approach would be to establish some sort of 'fast-track' system, whereby set-up of relevant trials could be expedited as necessary. Some interviewees envisaged such a system operating primarily where

a trial was already open at another hospital within a health board (e.g., a trial opened at a children's but not the associated adult hospital).

Other interviewees had more ambitious visions, proposing an 'idling agreement', activated only when an eligible and interested patient presented. However, these interviewees recognised that such a process would still require work from/in R&D Departments, as well as highly flexible and responsive research staff.

3.4 | Investing in the research workforce

Interviewees highlighted the diverse cast of people involved in setting up and delivering cancer trials: clinician-investigators, pharmacists, radiographers, pathologists, research support staff (nurses and data managers) and R&D staff. Interviewees perceived all these groups as working under considerable and often competing pressures, with these constraining professionals' engagement with trials in general, and TYA-relevant (rare disease) trials in particular (Hart et al., 2020). Interviewees suggested four ways in which investment in the research workforce might directly or indirectly facilitate the delivery of, and TYA's access to, relevant trials.

3.4.1 | Resourcing rare disease trials work appropriately

Interviewees drew attention to the particular resource issues raised by work on rare disease trials, intimating that with funding largely tied to recruitment, much of the work involved in trial set-up and maintenance went largely unrecognised. Some argued that new metrics were needed, involving some sort of weighting which recognised the difficulties and uncertainties of recruiting into trials targeting small populations, adding that the message from funders needed to be:

> '(We) know that you might only recruit one patient, but we want you to do it. So… we'll cover the cost.' (*DC-C-25*).

3.4.2 | Giving clinicians, pharmacists and radiographers ring-fenced time

Several interviewees made the case for investing in ring-fenced time which clinicians and other key professionals could dedicate to trials work:

> '(It's) really difficult when you're in a busy clinical job… small groups of investigators… have quite a huge portfolio of trials to manage, and there'll be nothing in their job-description that ring-fences time to do the regulatory bits… Investigators need to be given ring-fenced time.' (DC-C-26).

Fellowship schemes, where available, were described as enabling clinicians to deliver trials successfully and build (or sustain) their research profile—something interviewees viewed as essential if more lucrative commercial trials were to be attracted to their centres. Similarly, in those health boards and services where other pivotal staff had been given protected time (e.g., appointing dedicated clinical trials radiographers and/or 'backfilling' pharmacists' routine work), these investments were viewed as extremely valuable.

3.4.3 | Investing in research support staff (data managers and nurses)

Interviewees highlighted the benefits of re-distributing work often currently undertaken by consultants to increase capacity for trials work:

> 'The workload for the clinicians is dramatic, and they're all… very, very busy to start with… so to find a way for them to feel that they can, have more patients on clinical trials… however we can distribute the workload to do that… would be a benefit.' (DC-O-30).

Whilst some interviewees highlighted limits to the sort of work that could be delegated, support from data managers and research nurses with trial administration and recruitment was largely welcomed where this was available. Funding for research nurses at a network of centres which had committed to delivering a programme of haematology trials was cited by some as a very successful precedent: a similar investment in TYA-specific research support staff might 'really be practice changing' (DC-C-26).

3.4.4 | Supporting the development of research design and leadership skills

Interviewees mused that the pharmaceutical industry could not be relied upon to advance the care of small/minority patient groups or answer all the questions of interest to a publicly-funded healthcare system. They suggested that if the reach and breadth of trial portfolios were to be expanded, a larger cadre of 'enthusiastic individuals' (DC-C-13) with both the time and skills to conceive, design and establish trials was needed. Though not a quick fix, investing in leadership capacity was considered critical if the trials landscape was to be re-shaped. In light of the pressures on NHS staff, some interviewees envisaged clinical academics, employed partly by academic institutions, as playing a critical role here.

3.5 | Promoting pragmatism in trial design

Finally, to facilitate the engagement of professionals and successful recruitment of patients, interviewees highlighted the importance

of influencing the conception and development of trials, and advocating for pragmatic approaches to design. Proposals related to *encouraging more inclusive designs* and *considering the burden on participants*.

3.5.1 | Encouraging more inclusive designs

To maximise recruitment potential, some interviewees favoured opening trials with broader inclusion criteria, that is of relevance to a more diverse patient population. It was, however, noted that this was likely to mean opening studies with multiple strata and interventions. Having 'trials within a trial' might improve recruitment (and, where overarching approval was secured, bring about regulatory efficiencies) but these studies were also recognised as introducing new challenges, including unwieldy protocols which could prove difficult for direct care professionals to grasp and implement:

> 'The current trend is to have "umbrella" studies. And they can save time, passing lots of different studies through the regulatory authority… but essentially…. these are a collection of studies, under one mighty protocol, and that can be a bit of a job to manage.' (*DC-O-29*).

3.5.2 | Considering the burden on participants

Interviewees also stressed the importance of assessing, and minimising, the burdens on patients arising from trial participation, as these might act as disincentives to enrolment (and retention). To this end, some suggested reducing the number of data collection points:

> 'A lot of the trials – for our remote and rural population… there's too many time points of evaluation… we need to have a realistic approach to… the time points of assessment… and the paperwork… It's trying to kind of draw a bit of pragmatism into some of the trial designs.' (DC-C-17).

Additionally, the need for all information to be gathered face-to-face was questioned, with one interviewee suggesting that through more creative use of technology, 'you can… engage in different ways to get the same information' (DC-C-27).

Several interviewees emphasised the value and importance of getting input from patients at the design stage, rather than pre-supposing their perspectives:

> 'You have to make sure that the design of the trial takes into account the lifestyle of young people… PPI should be absolutely central to designing the trial, and not a token look at the information sheets at the end.' (DC-C-26).

These interviewees suggested that review and 'sense-checking' of draft protocols by engaged patients could substantially improve the success of recruitment.

4 | DISCUSSION

Interviewees suggested there was scope to address the challenges associated with delivering trials to small and dispersed populations and improve access to trials both for TYA with cancer *and* other patients with rare diseases. They portrayed this task as exacting but emphasised that the need to address barriers to delivering trials relevant to small populations was likely to grow. In support of this assertion, they drew attention to shifts towards targeted or 'personalised' treatment, explaining how this was, in effect, turning (sub-groups of) common cancers into 'rare' diseases. Interviewees suggested a wide range of ways research and care might be differently organised, administered and resourced, to facilitate increased trial access. Their proposals addressed both deterrents to trial sponsors/teams opening TYA-relevant trials more widely, and clinicians' (un)willingness to host such trials at their centres. Strategies were acknowledged as often presenting significant challenges and/or costs of their own.

The idea that the trials and treatment landscape are rapidly changing is echoed in the rare disease research literature. That too contends that, as the shift towards personalised/targeted medicine accelerates, the problem of how to successfully deliver trials relevant to very small patient groups will increase in significance. In 2015, (Bogaerts et al., 2015) reported that rare cancers accounted for almost a quarter of all cancer diagnoses. New ways of classifying (subsets) of common cancers (e.g., on the basis of molecular markers) mean the number of 'rare' cancers is rapidly growing (Billingham et al., 2016; Blay et al., 2016; Boyd et al., 2016). Care therefore needs to be taken not to construe 'rare' as intimating a minor/minority issue; effective strategies for organising and rewarding research and care relevant to small populations are becoming important to progressively larger numbers of patients and professionals.

In keeping with the literature on the challenges of delivering rare disease trials (Billingham et al., 2016), our interviewees stressed the importance of maximising the pool of potential recruits. Inevitably, the success (and perceived value of supporting) a trial hinges on the ability of investigators to enrol participants. To promote this, some interviewees advocated greater centralisation of services—a model under which all relevant patients would be referred to a designated centre for care and/or research. Paediatric oncology provides a precedent for this model; its high levels of trial accrual have been attributed to a more centralised approach to service organisation (Fern et al., 2014; Gatta et al., 2011). However, centralisation of services (generally) has historically proved contentious (Maybin, 2007) and our interviewees expressed reservations about referring (TYA) patients to more distant centres for care. For such a model to be acceptable, professionals' concerns about the mechanisms, administrative burden

and adequacy of practical—including financial—support for patients and families would need to be addressed.

Some interviewees argued for more flexible and collaborative approaches to trial delivery. However, though co-operative activity is less controversial and collaboration conceptually attractive (Ashley et al., 2015; Blay et al., 2016; Burke et al., 2007), such an approach was also considered problematic to engineer. Interviewees' concerns have some foundation: Weiss et al. (2015), for example, have warned that effective collaborations take considerable time to establish; changes to US research infrastructure, intended to promote collaboration, have been reported as having limited impact (Davis et al., 2017). In smaller countries, such as Scotland, where professionals have a history of working across organisational boundaries to provide routine services (and to a lesser extent support for trials), the foundations for collaboration may be firmer. However, interviewees' accounts suggest some challenges should still be anticipated, including resistance from (some) sponsors and/or principal investigators.

Our data additionally highlight the importance of streamlining the regulatory and administrative burdens associated with trial approval, set-up and maintenance, and managing their impact on the inclusion of rare disease studies in trial portfolios. Other authors have highlighted how cumbersome bureaucratic processes may (negatively) affect decision-making about whether to open TYA-relevant trials (Fern & Taylor, 2018; Gupta & Indelicato, 2014). Our own data offer some pointers to how activities might be streamlined, and burdens reduced, with this involving increased centralisation or standardisation of bureaucratic processes and practice(s). In the latter proposal, we see echoes of Crow et al. (2018), who, based on a review of delays in activating an international multi-centre trial, made the case for greater intra-national harmonisation of processes.

The value of investing in the research workforce was a further prominent theme, with interviewees highlighting the need to: ensure funding metrics/frameworks recognise the particular challenges of rare disease trials (rewarding activity as well as recruitment); resource the research time, and skill development, of a range of professionals. These proposals find support in the literature: Davis et al. (2017) have noted that rates of reimbursement for trials work are rarely sufficient to cover trial running costs and recommended increasing incentives for enrolment to trials where low rates of accrual are expected. Freyer and Seibel (2015) have identified increases in *local* resources as a particularly important facilitator of trial access, whilst Gupta and Indelicato (2014) and Fern and Taylor (2018) have advocated lobbying for more funding for infrastructure for the development and delivery of rare disease trials. However, even with additional funding, questions remain as to how capacity might be created. Workforce shortages in some services/areas may present considerable challenge to ambitions to give key staff 'ring-fenced' or research-focused roles: new ways of distributing work and diverting tasks from the staff groups under greatest pressure may also need to be found.

Fundamentally, the problem is a complex one and, as such, might require a sophisticated, multi-faceted, response. Accounts suggest that there are different ways, and levels, at which change might be instituted. Some of the proposals reported here would seem to be complementary (i.e. suitable to be undertaken in parallel) but not all. A proportion may be based on assumptions which have not been tested and might prove unworkable or ineffective.

4.1 | Strengths, limitations, future directions

Our inductive design and iterative approach allowed us to capture a diverse range of perspectives and draw out new and important findings. The proposals emerging from the study are grounded in participants' experiences of providing cancer care and delivering cancer research. Though the research was conducted in Scotland, the challenge of promoting access to trials for small and dispersed populations is in no way unique to that territory or the TYA patient group. This said, the relevance, and transferability, of our findings to other settings and populations is best judged on a case-by-case basis, by readers familiar with those contexts and groups. Finally, further work is needed to translate broad proposals emerging from our research into concrete, workable strategies. Real change is likely to require the support of a wide range of stakeholders, and consensus as to the best way forward. We suggest this might be achieved through one or more stakeholder priority setting exercises and close(r) scrutiny of favoured strategies.

5 | CONCLUSION

Re-configuring care, research, research administration and resource frameworks and priorities would—without question—have a price. However, the costs of doing nothing are also potentially significant. The first is the continued disadvantaging of, and discrimination in care offered to, patients with rare diseases, including (but not limited to) TYA with cancer. The second concerns the consequences for health systems of failure to adapt processes and infrastructure to reflect the changing focus of treatments and, therefore, trials. Avoidance of the challenge of making the delivery of rare disease trials viable may precipitate shifts in the global distribution of trials work, as the ability of some centres to deliver world class trials diminishes, and that of others, more ready to make the required changes, expands. If those health systems currently delivering complex trials are to remain important territories in trials research, establishing fit-for-purpose care, research and regulatory infrastructure, and investing more in the research workforce, including future research leaders, would seem essential. However, the support of a wide range of stakeholders is likely to be needed, if change is to have the desired effect.

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CONFLICT OF INTEREST

We have no conflicts of interest to declare.

AUTHORS' CONTRIBUTIONS

AJ conceived the idea for the study, which was designed and planned by JL and AJ, with input from RH. RH collected and managed the interview data. RH and JL conducted the analysis. RH prepared the first draft of this paper. All authors read and approved the final manuscript.

DATA AVAILABILITY STATEMENT

The data (interview transcripts) reported on in this paper are not publicly available as rendering them entirely un-identifiable would require significant redaction, and research participants did not consent to public data sharing. However, requests for access to the data underpinning this paper will be considered and – where reasonable – accommodated.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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