Project protocol

Project title

Transitions from paediatric to adult services for sickle cell disease (SCD): a prospective qualitative study examining young adult patients' experiences.

Summary of research

Transitions from paediatric to adult services cause problems worldwide, particularly among patients with chronic disease who have complex health needs requiring integrated care [13, 14]. Improvement of transitional care has become an important NHS strategic issue [25, 26]. Sickle Cell Disease (SCD) - a chronic debilitating condition that causes cumulative damage to multiple organ systems and acute pain episodes [15] - provides an excellent case study for examining transitions: it is complex and growing rapidly in the UK [16, 17], and there is a lack of UK-based research on this topic. In the UK, over 15,000 people live with SCD, 2/3 of them in London [18]. In England, it is the fastest-growing genetic blood disorder [17] with increasing associated mortality and morbidity [7]: between 2001/2 – 2009/10 SCD-related hospital admissions increased more than 50% [7], with the highest rates of emergency admissions among those who recently transitioned from paediatric to adult care, suggesting an urgent need for improvement in transitional care. Yet we lack empirical evidence about the support patients need during such transitions [29, 32]. Transition is a time of increased medical vulnerability and morbidity [29], and increased responsibility for self-care. Despite the importance of effective transitioning for managing SCD, many young adults reach the transition period without appropriate support [29]. This can have major negative consequences for them and for the health services, including missed appointments, poor treatment compliance and increased rates of emergency hospitalisations [22, 24], sometimes with life threatening consequences. For patients with SCD, prevention and recognition of complications are crucial to maintain health. Poorly-managed transitions have high economic and social costs to patients, their families and health services.

The paucity of UK data for any health condition on how social context mediates healthcare transition is a barrier to effective planning of support and delivering patient-centred transitional care [26]. Limited empirical research has explored the role of service provision components in achieving effective transitions but has failed to examine how these interact with patients' wider social contexts (such as how peer relationships, education and culture affect healthcare transitions) [3,8]. Our study will contribute to knowledge of this "neglected area" of care [25], for a marginalised condition [34,35] in the NHS and both focuses on the key NHS aim to provide 'patient-centred' care [18] and responds to recent calls for improvements in NHS transitional care [25,26].

Our research will bring together a unique group of multidisciplinary researchers (combining clinical, social science and adolescent health expertise), and will deliver value for money to patients and the NHS by contributing towards improving patient experience and care during healthcare transitions; care that will have a significant lifelong health, economic and social impact. Our aim is to improve services to reduce emergency hospitalisations, and improve health outcomes and quality of life, which will reduce both financial and also social and physical costs of SCD.

We use the case study of SCD to examine how NHS transitional care meets the holistic health needs of patients and of the support they need to move successfully into adult services [29, 40]. Analysis of one disease in depth will allow us to identify specific areas for improvement – including cost savings -- in SCD transition care, but will also allow better understanding of transitions generally thus

potentially expanding the cost-saving benefits of the project and the added value of improving quality of services and experience for a larger number of patients.

This qualitative research will examine the experiences of patients with SCD (13-21 years old) as they transition from paediatric to adult NHS services, analyse how their experiences are integrated into their whole lives (i.e. look beyond the clinical to other areas such as education and relationships) and obtain their perspectives on how transitions can be improved. The aim is to examine how transitions can be improved, with a strong focus on exploring patients' own experiences of the healthcare services they receive and taking into account their broader social context, such as education, and relationships.

Research questions: (Q1) What support and information resources are required by patients to improve transitional care? (Q2) What do patients perceive as the ideal process of healthcare transition? (Q3) How does healthcare transition affect young adults' healthcare-related behaviour (e.g. self-management) and quality of life over time? (Q4) How can patients' personal histories (past SCD crises and general health) enable or hinder transition success? (Q5) How does social context (e.g. carer relationships) affect transition success? (Q6) How can lessons from SCD be applied to other transitions?
We will collect and analyse data from patient interviews, patient audio diaries and case notes and provider interviews. We will recruit participants from a range of locations in two cities – London and another large English city (via SCD specialist healthcare services and from communities via our network of contacts).
Outputs from this research will include (but are not limited to): SCD transitional care support and information resources co-produced with patients; SCD transitional care digital story co-produced with patients to raise awareness about the findings; recommendations for provider training and support for improving transitions; analysis of how our findings can apply to other related chronic conditions (SCD comorbid factors or which share its symptoms and care needs) by identifying generic aspects of transition e.g. how patients' social contexts might shape their ability to move successfully into adult care.

Background and Rationale

Transitions from paediatric to adult services are often managed poorly resulting in significant costs [13,14]. Sir Ian Kennedy and the DH have called for improvements in NHS transitional care [25,26] and research is urgently needed to understand patients' needs and experiences during transitions [27]. SCD - a chronic condition that causes cumulative damage to organ systems and pain crises [15] - is an excellent case study for transitions because it is complex, under-researched, and places significant strain on the NHS, particularly in London [16]. SCD is the fastest-growing genetic blood disorder [17], affecting over 15000 people in the UK [18], with increasing associated mortality [7]. In England, SCD hospital admissions increased over 50% between 2001/02 and 2009/10 [7], with the highest rates amongst those having recently undergone transitions, suggesting there is room for improvement in SCD transitional care. Transitions are vital in SCD care because SCD patients become more vulnerable when they are transferred to adult services [7,20,21]. Poorly-managed transitions result in missed appointments, poor treatment compliance and increased rates of emergency hospitalisations [22,24] sometimes with life threatening consequences. For SCD patients, prevention and recognition of complications are crucial to maintain health. Our research has direct relevance to the NHS aim to ensure 'patient-centred' care [18]. It will be the first of its kind to examine what works and what could be improved in SCD transitional care from the perspective of NHS patients. Our findings will help improve transitions in linked conditions such as SCD comorbid factors [54, 55] or which share SCD care needs. Key elements such as how patients' social contexts shape their ability to move successfully into adult care and to self-manage own care to reduce health risks will be widely applicable.

The rapid growth of SCD and associated hospital admissions represent potentially avoidable economic, health and social costs for patients and the NHS [7, 8]. In 2010-2011 the total cost to the NHS of SCD-related hospital admissions was £16.2 million [8]. London absorbs 73.4% of all of England's hospital admission costs [8]. London accounts for the majority of the national SCD cases and SCD-related hospital admissions (74.9%), with 80% of all SCD-related admissions comprising patients from London's most deprived areas [7,28]. The distribution of SCD and its costs to the NHS makes London a good site for our research.

SCD NICE guidelines outline the importance of delivering culturally appropriate and person-centred care that treats patients as experts in their own condition [18]. This is particularly important during transition; a period when SCD patients are at risk of increased morbidity and mortality [29], when they experience complex developmental processes and when their adult health-related behaviour patterns develop [30]. Yet, transitional care is a "neglected area" of the NHS [25], with negative consequences for patients [13,14]. Unsuccessful SCD transitions can be devastating: SCD affects mainly black and minority ethnic groups who are more likely to suffer health inequalities [31,33]. SCD has for many years been marginalised within the NHS and UK society [34,35]. SCD patients are often poorly understood and stereotyped by healthcare professionals (e.g. as over-utilisers of opioids) [15,27]. Poorly managed transitions can increase the costs of SCD and the health inequalities that SCD patients in the UK face [36].

Patients with complex health needs, like those with SCD, find the experience of transition difficult and they require special support [30]. Sir Ian Kennedy [14] highlighted the need to ensure greater flexibility and continuity of transitional care. Transitions are decided on the basis of chronological age. For SCD patients whose age may not match cognitive development [33], this may mean difficulties in engaging in the active decision-making and self-care expected from patients in adult services [19]. There is no comprehensive evidence of how NHS transitional care meets the holistic health needs of SCD patients, despite the fact that 'patient experience' has acquired equal weight to 'patient safety' and 'clinical care' in quality assessment of NHS services [37,38], and despite responsiveness to patients' needs having recently been introduced as a patient experience indicator (2013 NHS outcomes framework [56]).

Our research will generate an evidence base on patients' experiences of transitional care, which can be used to improve care pathways and to empower patients in actively managing their own health. These improvements will help tackle the incidence of SCD-related illnesses and acute pain episodes [39]. Our findings will also provide information for other disease areas, where research has primarily explored the role of service provision components in achieving effective transitions but has failed to examine how these interact with patients' wider social contexts.

Research outputs that will benefit NHS patients and providers include: 1) SCD transitional care information resources to support patients during transitions and make the NHS principle of "no decision about me, without me" a reality, and 2) recommendations for providers so that they are equipped to meet patient needs and transitional care standards [18, 30].

Evidence explaining why this research is needed now

SCD patients are now living longer, yet little empirical attention has been paid to the support they need to move successfully into adult services [29,32]. The paucity of UK data for any health condition on how social context mediates healthcare transition is a barrier to effective planning of support and delivering patient-centred transitional care [26]. This knowledge gap can have devastating consequences and significant costs [13,14], particularly in London (where 2/3 of all SCD patients are located [7]). Many SCD patients lack appropriate support when transferred to adult services [29]. Yet from the limited evidence available we know that for other conditions, helping

patients achieve competence in areas of self-management and healthcare system navigation leads to more successful transitions [13].

SCD research has been dominated by a biomedical model that ignores the social dimension of the disease [41, 42], with correspondingly little exploration of how socio-cultural factors affect SCD healthcare transition [29]. Patients want transitional care to take into account their wider social context (e.g. religion) [43-45]: their context plays a key role in supporting or undermining how they cope with SCD [46]. Education and support must be based on an understanding of social context [47]. Research in other chronic conditions has emphasised the medical context but has not considered how patients' social contexts also affect healthcare transitions [3]. Most SCD transitions research has been conducted in the USA [42], and most studies have focused on service provision components rather than on patients' transition experiences [48]. SCD patients' experiences of transitional care have not been investigated in the UK. The social science approach we propose will make a distinctive contribution by complementing biomedical approaches [49,50]: a qualitative, social science focus is ideally suited for understanding how the social contexts in which patients live and receive care shape healthcare experiences [46].

Aims and objectives

This prospective qualitative research aims to explore healthcare transition journey from the perspective of young adult SCD patients in order to improve transitional care in the NHS more broadly.

There are six linked objectives:

(O1) to examine experiences of healthcare transition from the patient perspective (how transition occurs, healthcare services and support they receive and need during transition, skills they need).

(O2) to explore how current transitional care meets the holistic health needs of patients over time.

(O3) to understand patient concerns before, during and after transition and compare these with healthcare professional views on patient needs during transition.

(O4) to assess the quality of relationships and information exchange between patients and providers during transitions.

(O5) to develop resources to support SCD patients to move into adult care successfully and empower them to manage their own care.

(O6) to develop recommendations for providers to improve NHS transitional programmes and the care pathway of young adults.

Research questions

(Q1) What support resources are required by patients to improve transitional care? (O1,2,3,4,5,6).

(Q2) What do patients perceive as the ideal process of healthcare transition? (e.g. Are there areas where communication between patients and healthcare professionals during transitions can be improved?) (O1,2,3,4,5,6).

(Q3) How does healthcare transition affect young adults' healthcare-related behaviour (e.g. self-management) and quality of life over time? (e.g. Which are, in their view, the social and health costs that transitions have on themselves and their families? (O1,2,3,4,5,6).

(Q4) How can patients' personal histories (past SCD crises and general health) enable or hinder transition success? (O1,2,3,5,6).

(Q5) How does social context (e.g. carer relationships) affect transition success? (O1,O2,O3,O5,O6).

(Q6) How can lessons from SCD be applied to other transitions? (O6)

Methods

We use a qualitative design to examine how and why healthcare experiences change over time, patient experiences of transition and transition services [1]. A longitudinal component will help counteract recall problems and also prospectively capture patients' evolving experience of the unpredictable nature of SCD and of complex developmental processes [2], personal changes and changes in healthcare [3]. We will use mixed data collection methods to maximise analytical rigour [5] and will apply a triangulation protocol [6] to examine convergence, complementarity and dissonance of ideas across the data sources.

Sampling and setting

Participants will be purposively sampled from a range of locations in two cities – London and another large English city (via SCD specialist healthcare services and from communities via our network of contacts).

For SCD, London experiences are crucial. London has 2/3 all national SCD cases [7] and SCD-related hospital admissions' costs [8]. Insights into how patients' social contexts might shape their ability to move successfully into adult care will offer a new way to understand transitions and is likely to be relevant more generally.

Data collection

1. Longitudinal, one-to-one in-depth interviews with 20 patients in transition to adult care (10 aged 13-15 when the concept of transition is first introduced, 10 aged 16-18 at first interview, interviewing each again 6 and 12 months later).

2. In-depth interviews with a further 30 patients who transferred to adult services <2 years before interview (aged 19-21, once each) to obtain retrospective accounts of transition and current experiences of adult services. All in-depth interviews will explore Qs 1,2,3,4,5.

3. Audio diaries: 10 patients from each of the in-depth interviews groups asked to complete patient audio diaries (a non-intrusive way of collecting longitudinal data [9] about healthcare and social experiences during transition).(Q1,2,3,4,5).

4. Contemporaneous case notes for participants keeping audio diaries will be examined to compare the clinical records of their health status and treatment alongside their subjective diary and interview accounts of transition experiences (e.g. having many pain crises might have an important effect on transition experiences) (Q1,3,4).

5. In-depth interviews with 10 providers (paediatric/transition and adult services who have substantial involvement in the treatment of patient participants (e.g. lead paediatric, adult haematologist, nurse counsellor, nurse specialist, SCD social worker/clinical psychologist)), to examine providers' transitional care practices, perspectives and experiences, service changes over the research period (Q1,3,4,5).

6. Patient workshops: (one with ages 13-17 and one with ages 19-21) to co-produce support resources based on study findings, two further workshops (same two age groups) to co-produce the patient digital story.

We will conduct additional in-depth interviews in another large English city: 15 with patients in transition to adult care and five with service providers. We will also collect diary data from the 15 patients about their experiences during transition.

Data analysis

Analysis will follow principles of grounded theory [10] (systematic and comparative method for studying processes [11], particularly useful for studying the lived experiences of chronic illnesses [12]), and narrative analysis of individual accounts to examine the temporal ordering of events in transition trajectories, understand links between actions and consequences through time and across contexts.

Dissemination

This study will generate new evidence about patients' holistic health needs during transitions and the resources they need to move successfully into adult care. An original feature of the knowledge output is that it will not consider healthcare transition as a purely clinical experience. Rather, our sociological approach will situate this process within patients' broader social context.

The dissemination of findings and recommendations through advocacy and academic channels will be crucial in maximising impact. Crucially, through working in partnership with NIHR CLAHRC NWL our research findings will feed directly into a diverse stakeholder group who are actively involved in the improvement of SCD services in NWL through the development and implementation of evidence-based care pathways. This will ensure that research findings are rapidly translated into practice, with NIHR CLAHRC NWL's work acting as a template for implementation, which will be further disseminated through the national CLAHRC network and other routes including publication.

Patients and carers involved in the study will act as champions of the project. Building on their local and national networks we will work with them to engage SCD patients and healthcare professionals with the study findings and recommendations. We will work with our Universities' Press Office to engage the wider public with the research and use our project's twitter account to develop dialogue and gather feedback about the findings. We will involve young adults with SCD in co-creating a digital story using our findings and their own experiences to shape the narrative. The digital story will be used for advocacy work and awareness-raising among stakeholders and will be circulated to research participants together with the report of the study. The study outputs will be publicly available on our project blog. Findings will be disseminated through traditional academic channels: peer-reviewed publications (e.g. sociology and medical journals), conferences, widely accessed magazines (e.g. HSJ) and through our team's and SSC members' existing memberships (e.g. Imperial Paediatric Haemoglobinopathy Network, The Academic Paediatrics Association, Royal College of Paediatrics and Child Health, Clinical Reference Group for Haemoglobin) and international links (Muhimbili National Hospital SCD research team, Tanzania).

We will work with commissioners via existing links within our team. North West London commissioners are actively involved in the Early Years work of CLAHRC NWL.

Expected Output of Research

Outputs that will benefit NHS patients and providers within three years of project completion include:

(1) Downloadable SCD transitional care information resources for young adults to help them negotiate transition processes and manage their condition effectively produced in collaboration with and validated through a workshop with young adults with SCD.

(2) Co-produced SCD transitional care digital story to raise awareness amongst patients and healthcare professionals about the study findings and recommendations. Patient digital stories are

recommended for health promotion and advocacy, because of their power to engage diverse audiences and connect them around a common narrative [51,52].

(3) Evidence of patient perceptions and experiences of existing services which will feed directly into the design and implementation of evidence-based care pathways for SCD led by CLAHRC NWL and spread to other settings using the national CLAHRC network.

(4) Identification of aspects of transition care that need new thinking and redesign to meet patient needs. This will include recommendations for provider training and support for planning and managing transition care so they are better equipped to meet SCD patient needs and DH transitional care standards. USA-based research shows provider education and protocol development are crucial in providing quality transitional care [48]. This work will be translated into impact through partnership working with CLAHRC NWL to engage relevant stakeholders in commissioning and providing care and education.

(5) A comprehensive list of hypotheses about how the findings will apply to other related chronic conditions e.g. how the social contexts interact with service provision and shape patients' ability to successfully move into adult care and manage their own care. Whilst we are focusing on SCD, the knowledge and recommendation outputs will be transferable across the NHS and applicable to other conditions which are comorbid factors in SCD (asthma, depression, heart disease) or which share its symptoms and care needs (frequent service utilization, continuous self-management, e.g. Thalassaemia, congenital heart disease).

(6) Peer-reviewed journal articles.

(7) Articles in advocacy and healthcare provider publications.

(8) Conference presentations.

(9) Full report of the study (including an executive summary) detailing the research, findings and its policy, managerial and practice implications.

(10) Project blog/website and twitter account to disseminate lay information about the study.

Success will be assessed at the knowledge dissemination and knowledge translation level. Knowledge dissemination success will be assessed on the basis of our ability to engage stakeholders with the study findings and recommendations, measured by: N of publications and publication views/downloads; presentations at prestigious conferences; N of transitional care information output leaflets downloaded from study website and ordered directly from us; N of guidance and extent accessed of practical guidance for services on how findings can be translated into practice; N of followers and re-tweets from our project Twitter account; N of online views of our patient digital story; N of study website online visits.

Knowledge translation success measures based on: (1) evidence of effective collaboration with CLAHRC NWL to feed our research into the development of SCD care pathways CLAHRC NWL will conduct in parallel, (2) extent to which evidence generated though the study is adopted across the study sites and transferred to other NHS settings (a crucial step for long-term reduction in SCD patient health inequalities) and other conditions, (3) N of other long term conditions able to use our findings to improve service design and delivery e.g. asthma, allergies.

The potential risk of this longitudinal study is participant withdrawal (e.g. through poor health). To mitigate this, we will invest time and effort sustaining relationships with participants and maintaining their trust. The relevance of our findings to clinicians and their ability to be acted upon by service providers is a further risk. Working collaboratively with expert clinicians, experts in translation of research into practice and maintaining on-going dialogue with frontline healthcare providers and commissioners will mitigate this risk.

Approval by ethics committees

Important ethical considerations relate to issues of intrusion and confidentiality. The study involves asking participants at a vulnerable age to provide information of a personal nature. Key considerations include maintenance of confidentiality, and the adoption of a non-judgmental and respectful approach. We will follow National Children's Bureau ethical guidelines [57]. We will provide participants with age-appropriate written and verbal information about the study before they take part. 16-21 years old participants will sign a consent form prior to interview. We will gain assent from 13-15 years old but also obtain signed consent from their parent/carer. Before signing consent forms, we will ensure that they are fully informed and understand the nature and purpose of study. Participants will be assured that the study is confidential and entirely voluntary. For the 13-15 years old we will also explain that we will ensure confidentiality unless a child protection issue is disclosed [57], in which case we will report it via the relevant process. Interview, diary transcripts and patient case notes will be identified with a unique ID number, and the names corresponding with those numbers will be kept separately on a password-protected list (destroyed on completion of the project). No real names will be used in reports/publications and identifying details will be removed. We will provide a list of referral agencies to all participants should they need help with issues raised in interviews.

Ethical approval will be sought in parallel from the NHS Research Ethics Committee (NHS REC) and from the LSHTM ethics committee as soon as the project starts. Data collection will not start until ethical approval for the project has been obtained from both LSHTM and NHS REC review committees. For NHS REC the application will be made using the Integrated Research Application System (IRAS). Preparation of protocols for ethics, participant information sheet and consent form, and other supporting documentation for consideration during NHS and LSHTM ethical review will be a priority as soon as the project starts and will be finalised within the first three weeks of the project. We will involve patients in the development of participant information sheets and consent forms so that the information is as clear and understandable for research participants as possible. CM or AR will attend the meeting at which the REC will consider our application. A REC will give us ethical opinion on our application within 60 calendar days of receiving the application.

Patient and Public Involvement

This proposal has been developed in collaboration with patients. The idea originated from our interactions with patients and patient charities during our investigation into patient involvement in SCD healthcare improvement within NIHR CLAHRC NWL. Broken Silence (A charity organisation created by young adults with SCD) and Patrick Ojeer (father of SCD patient) have influenced the proposal throughout. We worked with them to identify research questions, examined their transition experiences and views on project’s aims and methods, all of which we incorporated. Patrick emphasised the aims should be to assess how transitional care enables patients to be treated according to the NHS ethos: 'No decision about me, without me' i.e. empower them to take responsibility for their care. Our objectives were formulated accordingly. Broken Silence highlighted the need to create tangible resources that would benefit patients, particularly to empower them. This influenced the idea of using research findings to co-produce support resources with young adults with SCD to help patients achieve competence in self-care and healthcare system navigation.

Patients will be actively involved in the following aspects of the research: design of the research, management of the research (e.g. study steering group), developing participant information resources, contributing to the reporting of the research and dissemination of research findings.

Broken Silence and Patrick Ojeer will play a key role in the management, leadership and design of the research through their Study Steering Committee membership, in which they will feed back on

study information sheets/consent forms, emerging findings and provide suggestions for analysis. We will involve them in producing reports and disseminating findings. Their involvement will be vital to ensure that patient perspectives shape the study. Both have a strong network of connections that we can utilise to disseminate study findings and engage patients with recommendations. The study's output resources will be co-produced with young adults with SCD. The Sickle Cell Society will be involved in disseminating findings. We will follow best practice guidelines for PPI developed by NIHR CLAHRC NWL. Patrick Ojeer (Sickle Cell Society Trustee, NHS England Specialised Commissioning Steering Group and Clinical Reference Group for Haemoglobin member) has already undergone training in the CLAHRC NWL Effective patient and community representative course and where applicable other members will be invited to participate. Independent advice and support will be provided by the CLAHRC NWL PPI team.

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