Supporting positive patient experiences for rare disease care during disruptive times: findings from a multicentre study

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Abstract

Introduction:

We describe the perceptions and experiences of health care services during the COVID-19 pandemic of those newly diagnosed with a rare, chronic inflammatory eye disorder.

Methods:

We undertook a cross-sectional study nested within a longitudinal multi-centre inception cohort study. Participants were families and young people (aged under 18 years) newly diagnosed with childhood uveitis. Using a validated tool, the Health Foundation COVID-19 Survey, we captured qualitative and quantitative data. Quantitative data were analysed using descriptive summary statistics. Qualitative, free text responses were analysed using content and thematic analysis.

Results:

Responses received from 60 families between 1st September 2020 and 30th March 2022 were analysed. Despite two in five reporting challenges in accessing medication, the majority felt comfortable accessing healthcare services (8%, 95% CI 3 - 18%, of participants expressed discomfort, versus 28%, 95% CI 26 – 28% of general population). Thematic analysis identified five themes: the value of protected spaces to safely access care; the positive role of digital health tools the experience of immature telemedicine; disintegration of care; and dealing with uncertainty.

Discussion:

Our findings suggest that families of children with a rare chronic condition had greater confidence in accessing healthcare during the pandemic than the general population. Nevertheless, to ensure more robust health services for such populations in future times of disruption, developments in telemedicine should be directly informed by the experiences of those with rare disease. The development of new healthcare processes which ensure the whole healthcare team has adequate information to support families should be prioritised.

What was known before

- Eye health care service planning is typically informed by the experience of a patient population predominantly comprising older people, with common disorders, many of which are associated with aging.
- To be appropriate, development of robust services should be informed by the voices of two groups who are often overlooked: namely children, who comprise 1 in 5 of the UK population, and those with rare disease, who collectively comprise 1 in 17 individuals.

What this study adds:

- During times of disruption, safety measures, facilitative digital tools and integrated patient care pathways support patient reported positive rare disease experiences.
- Uncertainty, known to typify rare disease journeys, is a central theme in negative experiences.
- Care processes which support integrated care in disruptive times, and which empower parents to be advocates for their child's care, should be prioritised.

INTRODUCTION

The COVID-19 pandemic disrupted all elements of global health care services. Lessons learnt from positive and negative patient experiences during this disruptive period, and the service-specific factors associated with those experiences, have been disseminated across care settings. This provides evidence to develop more robust services ahead of the further periods of healthcare disruption predicted by many.[12] Health service planning is typically informed by a patient population predominantly comprising older people, with common disorders, many of which are associated with aging. To be appropriate, redevelopment of robust services should be informed by the voices of two groups who are often overlooked: namely children, who, for example, comprise 1 in 5 of the UK population,[3] and those with rare disease, who collectively comprise 1 in 17 individuals.[4]

Uveitis encompasses a family of rare disorders characterised by intraocular inflammation with or without accompanying systemic disease.[5] Uveitis of childhood onset is further characterised by chronicity,[6] frequent contact with health services,[6] significant (visual and quality of life) morbidity,[7] and the need for immunosuppressive treatment and highly specialised multidisciplinary care.[5,68] Children are usually diagnosed in secondary or tertiary care, may require input from quaternary care centres,[5,6,8] use pharmacy or blood testing services in the community,[6] depend on other primary care services for co-ordination of care, and need hospital emergency services for disease flares or complications of immunosuppression.[6,7] These children and their families are well placed to describe experiences of interacting with and between different levels of healthcare. This study aimed to describe and explore the perceptions and experiences of healthcare service use by those children newly diagnosed in the UK with uveitis during the time of significant disruption caused by the global COVID-19 pandemic.

SUBJECTS AND METHODS

We undertook a mixed-methods cross-sectional study embedded within the Uveitis in Childhood Prospective National Cohort Study (UNICORNS).[9] This is a multicentre inception cohort study recruiting children newly diagnosed with non-infectious uveitis since March 2020, with recruitment active across 32 National Health Service (NHS) Hospitals.

Study participants

Families of children aged up to 18 years and newly diagnosed with non-infectious uveitis were invited to participate.

Data collection tool

The Health Foundation COVID-19 Survey was selected for use as a tool developed, piloted, and validated by the Health Foundation, (a health policy non-governmental organisation)[10] to evaluate the effect of the pandemic on the perceptions and experiences of health service use by members of the UK public. Its use therefore also allows comparison with the general UK population. The first section measures the impact on health and wellbeing, and the second measures experiences of using NHS services and perceptions of how services are managing. The survey allowed a convergent mixed-methods design, with concomitant collection of quantitative (closed text responses) and qualitative (free text responses) data. The survey was adapted for use in this study (Supplement S1), with respondents being asked about experiences and perceptions of health care use for their child's disorder(s) during the time around diagnosis.

Survey administration and data collection

The questionnaire was sent via post to all eligible participants following recruitment into the UNICORN study. Children aged older than 14 years were asked to complete the survey themselves, whilst for younger children the survey was completed by the parent / carer. A member of the study team (SK) manually entered responses into study databases with entries reviewed by a second investigator (ALS).

Analysis

Quantitative data from closed ended questions were analysed using descriptive summary statistics. Qualitative analysis was then applied to the responses to open ended questions in the survey. Firstly, content analysis[11] was undertaken on responses to open ended questions on the direct impact of the pandemic on health and wellbeing. In order to explore patterns and themes around the perceptions and experiences of health care use, thematic analysis[12] was undertaken on free text responses to those questions. Two investigators (SK, a non-clinical research assistant, ALS, a researcher and senior paediatric ophthalmologist) initially immersed themselves with all qualitative responses to gain understanding of the content and attitudes present. Qualitative analyses were undertaken using Microsoft excel with colour coding labels.

For the content analysis of responses on the 'direct impact of the pandemic on health and wellbeing', two coders (ALS, SK) worked together in an iterative, negotiated process to develop a coding system to summarise the data. Following the drawing of initial conclusions around key categories of data, data were re-interrogated to identify any characteristics not captured within those categories.

The thematic analysis of ‘perceptions and experiences of healthcare service use’ was undertaken using a hybrid of deductive and inductive approach, whereby the data were coded and categorised deductively as ‘positive’ or ‘negative’ experience, and by generating new codes inductively to capture the issues emerging spontaneously from the data. This approach was used to condense data into meaningful categories and applied iteratively to develop and label the key overarching themes.

Themes were generated on a non-exclusionary basis, with some participants’ comments able to provide data which belonged to more than one theme. All steps of the analysis were conducted independently by both researchers with peer discussion on completion of every step. Initial codes for labelling the data and emergent themes were identified independently (ALS, SK) and then compared and agreed through group discussion (SK, ALS, VT). The resultant overarching themes were then reviewed to examine fit with the data, and to identify any characteristics not captured within the thematic map, with themes revised or relabelled as needed.

Patient and public involvement

Our patient research partner group, the Childhood Uveitis Study Steering Group (established in 2019 to support the UNICORN study)[13] was involved in the study’s design, conduct, and interpretation of the data.

RESULTS

Of 113 families first approached to take part in the survey, 53% responded. The analysis therefore included data from sixty respondents (45 parents or carers, 15 young people) representing uveitis cases diagnosed between 1st March 2020 and 20th November 2021. Most participating families were from a white ethnic background, with over-representation (relative to national demographics) of those owning their homes or having two or more cars (Table 1).

Table 1
Characteristics of participants

	C-19 adult, n = 45 **% (n)**	**C-19 Child, n = 15** **% (n)**	**2021 UK general population** **%[14]**
Number of patients			
Age group of respondent			
Less than 20 years	0	100 (15)	-
21 to 30 years	2.2(1)	-	-
31 to 40 years	37.8(17)	-	-
41 to 50 years	46.7(21)	-	-
More than 51 years	13.3(6)	-	-
Ethnicity of respondent			
White - English, Scottish, Welsh, Irish	60 (27)	60 (9)	76
White other	16 (7)	20 (3)	6
Black African	2 (1)	0	3
Indian	2 (1)	0	3
Pakistani	7 (3)	0	3
Bangladeshi	2 (1)	0	1
Chinese	0	0	1
Asian other	2 (1)	0	2
Other	9 (4)	20 (3)	5
Gender of respondent			
Female	87 (39)	87 (13)	51
Family structure			
Two parent family	91 (41)	87 (13)	83
Single parent family	7 (3)	7 (1)	15
Other	2 (1)	7 (1)	2
Car ownership of parents			
None	4 (2)	20 (3)	32

	C-19 adult, n = 45 % (n)	C-19 Child, n = 15 % (n)	2021 UK general population %[14]
One	27 (12)	33 (5)	48
Two or more	62 (28)	47 (7)	20
Home ownership status of parents			
Owned	76 (34)	87 (13)	63
Privately rented	11 (5)	0	20
Rented from a housing association or local authority	11 (5)	13 (2)	17
Other	2 (1)	0	-

Impacts of the pandemic on health and wellbeing

The majority of respondents expressed concerns over the direct or indirect impact of the pandemic (Table 2), and more than half reported difficulty accessing basic food or household items. Compared to the general public,[15] a higher proportion of responding families reported challenges in accessing essential medication, and in communicating with friends and family (Table 2).

Table 2
Impact of pandemic on well-being and daily life of families

Area of concern for n = 45 parent responders	**Fairly concerned** **% (n)**	**Very concerned** **% (n)**	**Total expressing concern** **%,** **(95% CI)**	**UK general public levels of expressed concern¥** **%,** **(95%CI)[15]**
Risk posed to personal / family health & wellbeing	47 (21)	22 (10)	69 (53–81%)	55 (53–57%)
Risk of knock-on impact to personal / family health & wellbeing	56 (25)	16 (7)	71 (56–84%)	73 (71–75%)
Risk posed to health & wellbeing of the nation	58 (26)	27 (12)	84 (71–94%)	86 (84–87%)
Risk of knock-on impact to the health & wellbeing of the nation	64 (29)	24 (11)	89 (76–96%)	94 (93–95%)
	A little harder % (n)	Much harder % (n)	Total %, (95% CI)	UK general public %, (95% CI)
Communicating with friends/family	56 (25)	9 (4)	64 (49 - 78%)	47 (45–49%)
Being able to get essential medication	31 (14)	11 (5)	42 (28–58%)	24 (22–26%) [a]
Being able to get basic food items	58 (26)	9 (4)	67 (51–80%)	54 (52–56%)
Accessing green space locally	27 (12)	2 (1)	29 (16–44%)	33 (31–35%)
Being able to get household goods	58 (26)	4 (2)	62 (47–76%)	49 (47–51%)

¥From the Health Foundation Wave 1 survey of 2102 adults, November–December 2021

[a] Difference in proportions z score 2.8, $p < 0.01$

Content analysis of open-ended responses revealed seven categories of concern around the health of the respondent and their family, specifically: the direct risk of contracting COVID-19, the lack of exercise, closure of leisure activities and weight gain. There were concerns that their child was put at additional risk due to their diagnoses. *“Due to A's immune system being affected…we are concerned for A’s health if A was to come into contact with the virus”.* Worries over the impact on mental health were described by many: *‘My main concern about the virus involves the impact of lockdowns and restrictions on mental health’*. Families shared treatment specific concerns around the negative outcomes of immunosuppression, perceiving an increased risk of contracting SARS-Cov2, and increased risk of poorer outcomes following COVID infection: *“With B’s immune system compromised due to the medication it’s scary”; “we are fearful C could get more ill or may not be able to fight virus if infected…”; “..D is on methotrexate & I'm very concerned that it's…easier to catch the virus…”.*

The final category of concern was the financial impact on well-being, with expectations of negative national economic impact, worsening public services, rises in the cost of living, and job losses or poor job security, reported by the majority of respondents.

Perceptions around, and experience of, using NHS services

Most families described feeling comfortable when using NHS services (Table 3) during the pandemic for the management of newly diagnosed uveitis, with a lower proportion expressing discomfort with the use of local hospitals, or of accident and emergency departments when compared to the general population.

Table 3
Reported levels of discomfort on using health services

Health Service	**Quite uncomfortable** **% (n)**	**Very uncomfortable** **% (n)**	**Total expressing discomfort** **%,** **(95% CI)**	**UK general public levels of expressed discomfort**[¥] **%,** **(95% CI)** [10]
GP practice (used by n = 55)	13 (7)	11 (6)	24 (13–37%)	15 (13–17)
Local hospital for a non-emergency or routine appointment (used by n = 56)	9 (5)	0 (0)	9 (3–20%)	22 (20–24%)[a]
NHS 111 telephone services (used by n = 44)	2 (1)	5 (2)	7 (1–19%)	10 (9–11%)
Pharmacies (used by all respondents)	5 (3)	0 (0)	5 (1–14%)	14 (13–16%)
Accident and emergency at hospital (used by all respondents)	5 (3)	3 (2)	8 (3–18%)	28 (26–30%)[b]

[¥] From the Health Foundation Wave 1 survey of 2102 adults, November–December 2021

[a] Difference in proportions z score − 2.4, $p < 0.05$

[b] Difference in proportions z score − 3.3, $p < 0.01$

Thematic analysis of free text responses on perceptions and experiences of health care service use, provided in section two of the survey, identified five themes, of which two captured 'positive' (protected spaces and digital health tools), and three 'negative' experiences and perceptions (telemedicine immaturity, fragmentation of care, and the unknown) (Fig. 1).

Positive experiences and perceptions

Protected spaces

Many families and young people expressed gratitude for the precautions taken within hospital based clinical areas, with visible and consistent adoption of safety measures conferring a sense of being protected at a vulnerable time. The phrase ‘safe’ was used multiple times when respondents were describing positive perceptions on their health service environment:

Everyone is taking the right precautions to make the appointments covid safe....

….precautions have been taken to prevent a spread of the coronavirus in the hospital.

Everyone we've encountered has been helpful, calm, informative and covid safe….(YP, response from young person)

a lot of safety measures were taken so I didn't worry about catching COVID while I was there (YP)

They {doctors and nurses}…make me feel safe (YP)

These precautions were, for those who had never before experienced specialist hospital-based services, a positive introduction to spaces which they knew would form a long term ‘home’ for the care of their chronic condition:

Didn't want to go but trust the people that work there (YP)

“*….anyway, the risk of not being seen would be greater*”.

Adoption of digital health tools

Families described positive experience of the adoption of telemedicine and digital health tools (e.g., particularly those supporting synchronous and asynchronous care) across all levels of care from primary care services to quaternary care services:

*xxx {*digital health application, DHA*} service had been very goo*d

*We contact the GP using xxx {*DHA*} which works well*

*We’ve been using xxx {*DHA*} to contact the hospital team and get responses*

blending face to face appointments with virtual appointments….diagnosis, treatment and care were not compromised

A much-mentioned benefit was the ability to share patient generated image data with clinical teams:

….send photos if necessary

“*….easy to send photos*”.

Negative experiences and perceptions

Telemedicine immaturity

The absence of mature telemedicine systems underpinned many negative experience and perceptions. System immaturity was reflected in multiple ways, including: reliance on voice telephone contact only due to the absence of video communication, and systems which had not yet been refined or informed by patient experience and were therefore overly complicated for families to navigate:

Telephone assessments….getting seen by GP only telephone was a worry

…. frustrated by the complicated appointments system

App used for triage makes it more complicated to get a phone appointment..

the appointments system [is]*…not currently fit for purpose*

[secondary care centre]*….did not ask to see daughter's eyes…. diagnosis + treatment were delayed*

Fragmentation of care

The negative impact of the perceived failures in co-ordinating care was reported by families and young people, with negative experience of synchronous consultations with primary and secondary care teams without access to clinical information from specialist centres.

….told different things about our medicines by GP, pharmacy and local hospital

*{*local*} hospital….did not take our concerns seriously and didn't have any knowledge about condition*

Families also reported that primary care teams were often unaware of the urgency or severity of the potential need of their child for care, despite the families attempt to communicate this:

*….told no hospital referrals {*in*} local area at present only urgent referrals*

….seem to fob you off

….they did not listen to me….

Young people in particular reported negative experiences of perceived gaps in the co-ordination of care, expressing concerns around the awareness of their primary and secondary care teams of particular needs of their disorder:

When I attended the xxx hospital I am not confident with them (YP)

…this makes me nervous to see xx again as I don't think xx knows what xx is doing (YP)

…didn't really listen as much…(YP)

The unknown

The perception that they, their family, the country and the world were navigating uncharted space emerged frequently across responses from parents and young people and across different care settings. Families and children described not knowing how to judge their direct risk of contracting COVID-19, the impact of the use of immunosuppression, how their clinical course and access to care would be disrupted by the pandemic, and the risk to their wider communities. This was often anchored in their understanding of the potential uncertainties ahead with their child's journey through their first years following diagnosis of a complex rare disease.

…new virus and {I'm} nervous…(YP)

*Because nobody knows the {*coronavirus*} side effects*

DISCUSSION

From this mixed methods study embedded in a national cohort, we report that families of children newly diagnosed with a potentially disabling chronic inflammatory eye disease during a time of considerable disruption maintained confidence in the UK National Health Service. They expressed a low level of discomfort in using those services despite concerns around the wider impact of the pandemic. Families' accounts of their experiences included positive experiences of protected spaces and informed clinicians and digital health tool implementation. However, there were negative perceptions around the impact of telemedicine immaturity on care services alongside perceived or actual failures in the co-ordination of care. The fear of the unknown was expressed by many, with the uncertainty of the rare disease journey ahead compounded by the uncertainties brought by the pandemic.

Our study is strengthened by the use of a nested approach which took advantage of an already underway inception cohort study. The study provided a contemporaneous insight into perceptions and experiences for UK patients and families embarking on a journey of multi-system, multi-level care for a rare disease. Study limitations include a small sample size and the likelihood of some elements of response bias. Families who are particularly vulnerable to negative experiences of health care service, specifically those from socioeconomically deprived or non-White backgrounds,[16,17] are under-represented within the respondent sample compared to the overall UK population. There was an overall decline in population mental health during the pandemic,[18] attributed to the distinctive circumstances of societal lockdown related isolation, acute and acute-on-chronic financial strain, concerns around chronic health conditions [19], and changes in household dynamics.[20] This decline disproportionately affected people from a lower socioeconomic background.[18] The experience and perceptions of these families may be less positive, and grounded in different domains, with different lessons which need to be learned in order to develop equitable health care service provision during disruptive times. The sociodemographic health care access and provision inequities starkly highlighted by the pandemic[21,22] may have resulted in inequitable experiences of health services during the pandemic, which may not be captured by our survey findings because of the under-representation of families from more vulnerable socioeconomic strata. Another key limitation was the absence of direct exploration of the determinants of family or care-giver mental health and well-being or their pandemic experiences through in-depth interviews. Whilst the population for this study were affected by eye disease, families did not report specific themes around the additional impact of visual impairment on pandemic experiences. The study is therefore unable to explore the intersectional impact of accessibility concerns on the experiences of health care use. Additionally, the study used a survey tool which has not yet been validated for use in those aged under 18 years, with data from young people and parents/carers considered in combination.[15] Future work may benefit from the collection of data only from children themselves, to gain a stronger sense of their perspective. Despite these limitations, the findings from our unique cohort show the value of our pragmatic approach. Using free text responses to add nuance to survey collected data, this study has generated useful evidence, informed by the experiences of families with rare disease, for service redevelopment.

The adoption of face masks, protective personal equipment (PPE) and social distancing across hospital services was recognised and welcomed by families. Our findings on the feelings of safety expressed by families attending hospital appointments may offer vulnerable patients reassurance and encourage attendance during future pandemics. Whilst the adoption of digital interventions during the pandemic was perceived as supporting some aspects of care, with health tools allowing families to share media or request actions or information directly from care providers, other aspects of care were not well served. Platforms used to organise and deliver telephone-based consultations, and the absence of video-based consultations to replace face to face appointments were perceived poorly by families seeking primary health care services for their child. The additional information conveyed through visual communication may be necessary to facilitate understanding for discussions around complex or rare disorders. Although the provision of remote primary care consultations has found some favour with the UK public,[23] our findings suggest that pathways enabling synchronous and asynchronous conveyance of visual information (eg, physical appearance or behaviours) should be prioritised for families seeking care for rare or complex conditions. Additionally, the burden of rare disease often disproportionately falls on those families from socioeconomically and educationally disadvantaged backgrounds, those with limited or no access to the internet, or whose first language is not English.[2425–27] Although quaternary care for patients with rare diseases is delivered in specialized centres, the need to devolve safely and effectively certain aspects of such multidisciplinary care to local providers such as general practitioners or community pharmacists remains.

Our findings also suggest that primary and secondary care teams require additional support in delivering and co-ordinating care for those with rare disease. Managing the fear of the "unknown" will necessitate sharing clinical experience and promoting further research. Answers emerging through these processes will need to be appropriately communicated to patients and their families in order to best support patients and avoid science misinformation and disbelief. While further maturation of information and communication technologies is probably a matter of time, the responses of patients and their caregivers in our cohort emphasizes the core need to ensure optimal communication among healthcare providers. For this reason, it will be important to devise specific patient educational activities through the coordinated involvement of healthcare professionals and dedicated patient groups. A key example from this cohort is the messaging around the protection of children on immunosuppressive agents. Early in the course of the COVID-19 pandemic, concerns around the risk SARS-COV-2 posed to immunocompromised individuals led to advice on ‘shielding’ those individuals from contact with others to avoid viral transmission. Later on, evidence emerged on the reduced risk of adverse outcomes following COVID-19 infection for children, even those on immunosuppressive therapies.[28 29,3031]. While dissemination of this information amongst specialists prescribing immunosuppression to children was timely, this may not have been the case for primary care providers, with general practitioners, pharmacists and patients appearing to give and receive conflicting advice. One approach to address this challenge will be empowering parents/carers to use their expertise to as advocates for the care of the child, and for health professionals to recognise that parents are ‘experts’ in many aspects of their child’s care.

The need to restructure clinical pathways in response to the pandemic created opportunities to test the feasibility of novel approaches in healthcare provision. These approaches could help increase throughput while dealing with clinical backlog in the aftermath of the pandemic, and serve as a template for dealing with future public health emergencies, with particular reference to the integration of primary and specialised care services. The exceptional circumstances of the pandemic have brought into stark relief the elements of healthcare which require uninterrupted service, and it is the apparently ‘outlying’ populations which may carry exemplars of best practice, or barriers to achieving good health outcomes. Our findings from this rare disease cohort suggest that these patients should have prioritised access to visual based communication with care providers, and that sources of information should be clearly signposted to patients and community care providers. Additionally, families need to be empowered to be advocates for their child’s care e.g., through the use of parent information strategies.[32] These suggestions should inform and support the ongoing post-pandemic redevelopment of care.

Declarations

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Conflict of Interest

There are no disclosures to report.

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Ethics

Ethics approval was granted by a UK Research Ethics Committee (reference 20/LO/0661 Bloomsbury Research Ethics Committee) and written informed consent was obtained. Study registration number: NCT05385757 (ClinicalTrials.gov)

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Figures

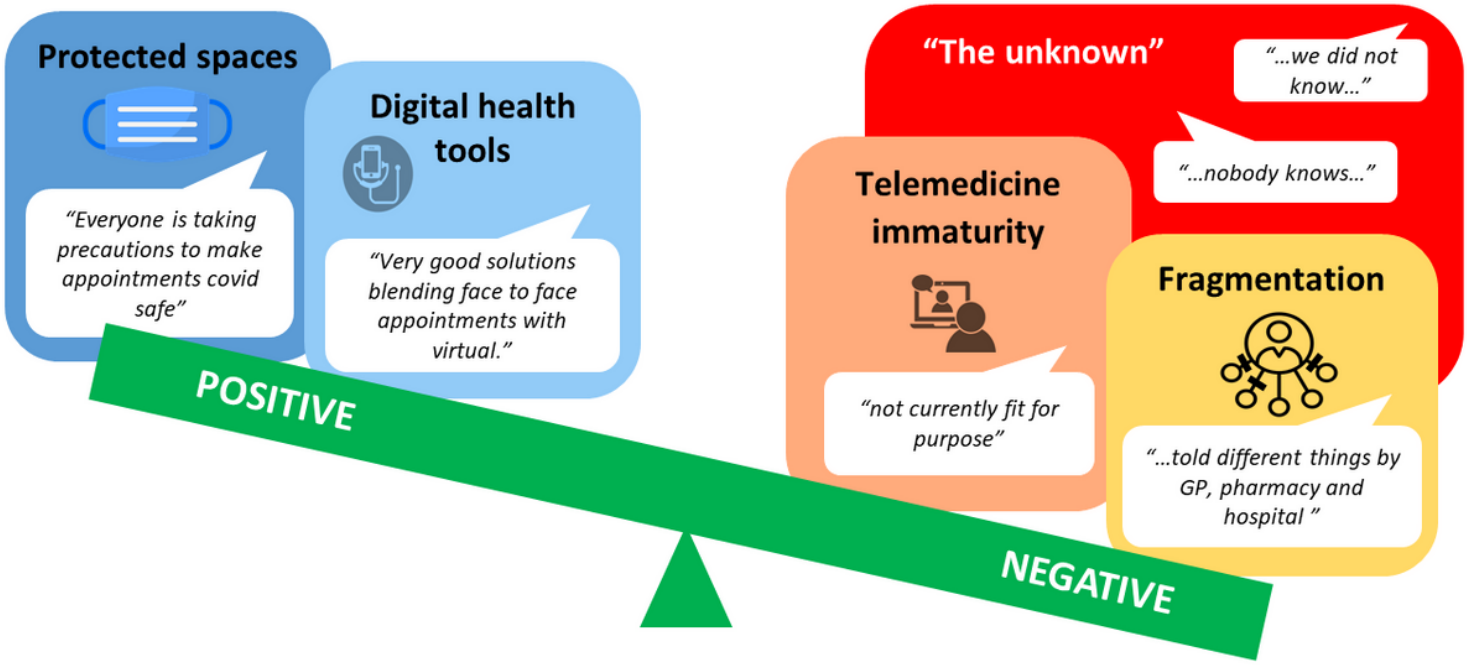

Figure 1

Themes around the positive and negative perceptions and experiences of health care service use

Supplementary Files

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