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Isolation, Uncertainty and Treatment Delays: Parents' Experiences of Having a Baby with Cleft Lip/Palate During the Covid-19 Pandemic

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Manuscript ID	CPCJ-21-0340.R1
Manuscript Type:	Original Article
Keywords:	Parental perception, Psychosocial adjustment, Quality of life
Abstract:	<p>Objectives: Previous literature finds that having a child with a cleft lip and/or palate (CL/P) may pose social and emotional challenges for parents. For parents of children born during the Covid-19 pandemic, such challenges may be heightened. Further, novel demands brought about by the pandemic could have caused additional hardships. The aim of this study was to describe the impact of the pandemic on new parents through qualitative exploration of their experiences.</p> <p>Design: Semi-structured interviews were conducted with 14 parents of children born in the United Kingdom with CL/P between January and June 2020, around the start of the pandemic. Data were analysed using inductive thematic analysis.</p> <p>Results: Three themes, with sub-themes, were identified. The first theme, "Changes to Healthcare: The Impact of Restrictions and Reduced Contact", discussed the impact of the pandemic on perinatal care, the care received from the specialist CL/P teams, and parents' experiences of virtual consultations. The second theme, "Family Functioning During the Pandemic", covered parental anxiety, fathers' experiences, and social support. The third theme, "Surgical Prioritisation: Delays and Uncertainty", addressed changes to surgical protocols, coping with uncertainty, complications associated with delayed surgery, and how parents created positive meaning from this period.</p> <p>Conclusions: A range of increased and additional psychosocial impacts for parents were identified, along with several coping strategies, utilization of social support, and the positive aspects of their experiences. As the pandemic continues, close monitoring of families affected by CL/P remains imperative, particularly for those at risk of emotional distress.</p>

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Table 1. Overview of Interview Guide	
Demographic information	<ul style="list-style-type: none">• Parent’s date of birth• Parent’s ethnicity• Parent’s marital status• Parent’s employment status• Child’s date of birth• Child’s ethnicity• Child’s cleft type• Cleft team• Family history (e.g. other children, known history of CL/P)
Early experiences	<ul style="list-style-type: none">• Experiences of pregnancy• Experiences of diagnosis• Experiences of birth and postnatal care• Introduction to specialist cleft nurse and cleft team
Adjustment and wellbeing	<ul style="list-style-type: none">• Adjustment after birth• Positive experiences with baby
Information and support	<ul style="list-style-type: none">• Sources of information• Types of information• Support from friends and family• Support from specialist healthcare professionals• Support from CLAPA• Evaluation of support/information received (positive, negative, amount, areas for improvement)
Treatment	<ul style="list-style-type: none">• Changes/delays to child’s treatment• Communication of changes by cleft team• Involvement in decision making about child’s treatment• Concerns around changes/delays• Family attempts to mitigate anticipated impacts of changes/delays• Evaluation of support through changes/delays• Positive outcomes of changes/delays
The future	<ul style="list-style-type: none">• Future treatment plans• Covid-19 protocols for treatment• Concerns about future• Hopes for future

Table 2. Demographic Information

Participants			
	Mother	Fathers	UK census data^a
Parent	71.4% (<i>n</i> = 10)	28.6% (<i>n</i> = 4)	
Age in years (mean and SD) ^c	31.90 (2.685)	33.25 (4.193)	
Ethnicity			
White British	80% (<i>n</i> = 8)	100% (<i>n</i> = 4)	80.5%
Other	20% (<i>n</i> = 2)	0% (<i>n</i> = 0)	19.5%
Marital status			
Married	50% (<i>n</i> = 5)	50% (<i>n</i> = 2)	47%
Engaged / Cohabiting / Long term relationship	30% (<i>n</i> = 3)	50% (<i>n</i> = 2)	-
Single	20% (<i>n</i> = 2)	0% (<i>n</i> = 0)	35%
Employment status prior to birth			
Full time	70% (<i>n</i> = 8)	100% (<i>n</i> = 4)	
Part time	20% (<i>n</i> = 2)	0% (<i>n</i> = 0)	
Maternity/paternity leave at time of interview			
Yes	100% (<i>n</i> = 10)	0% (<i>n</i> = 0)	
No	0% (<i>n</i> = 0)	100% (<i>n</i> = 4)	
Immediate family history of cleft			
Yes	0% (<i>n</i> = 0)	0% (<i>n</i> = 0)	
No	0% (<i>n</i> = 0)	0% (<i>n</i> = 0)	
First child			
Yes	50% (<i>n</i> = 5)	75% (<i>n</i> = 3)	
No	50% (<i>n</i> = 5)	25% (<i>n</i> = 1)	
Participants' Children^d			
			CRANE data^b
Age in months (mean and SD) ^c	5.30 (1.703)		
Gender			
Male	80.0% (<i>n</i> = 8)		
Female	20.0% (<i>n</i> = 2)		
Presence of syndrome			
Yes	0.0% (<i>n</i> = 0)		
No	100.0% (<i>n</i> = 10)		
Cleft type			CRANE data^b
UCLP	40.0% (<i>n</i> = 4)		21.7%

BCLP	10.0% (<i>n</i> = 1)	9.6%
UCL	40.0% (<i>n</i> = 4)	25.3%
BCL	0.0% (<i>n</i> = 0)	
CP	10.0% (<i>n</i> = 1)	43.5%
^a Office for National Statistics (2018)		
^b Cleft Registry and Audit Network (CRANE; 2020)		
^c Ages correct in September 2020		
^d Note that although there were 14 parents, there were only 10 children as there were 4 parent couples (i.e., mother and father) who took part		

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Cleft Lip/Palate During the Covid-19 Pandemic

Abstract

Objectives: Previous literature finds that having a child with a cleft lip and/or palate (CL/P) may pose social and emotional challenges for parents. For parents of children born during the Covid-19 pandemic, such challenges may be heightened. Further, novel demands brought about by the pandemic could have caused additional hardships. The aim of this study was to describe the impact of the pandemic on new parents through qualitative exploration of their experiences.

Design: Semi-structured interviews were conducted with 14 parents of children born in the United Kingdom with CL/P between January and June 2020, around the start of the pandemic. Data were analysed using inductive thematic analysis.

Results: Three themes, with sub-themes, were identified. The first theme, "Changes to Healthcare: The Impact of Restrictions and Reduced Contact", discussed the impact of the pandemic on perinatal care, the care received from the specialist CL/P teams, and parents' experiences of virtual consultations. The second theme, "Family Functioning During the Pandemic", covered parental anxiety, fathers' experiences, and social support. The third theme, "Surgical Prioritisation: Delays and Uncertainty", addressed changes to surgical protocols, coping with uncertainty, complications associated with delayed surgery, and how parents created positive meaning from this period.

Conclusions: A range of increased and additional psychosocial impacts for parents were identified, along with several coping strategies, utilization of social support, and the positive aspects of their experiences. As the pandemic continues, close monitoring of families affected by CL/P remains imperative, particularly for those at risk of emotional distress.

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Key words: cleft lip and palate, parents, Covid-19, surgical prioritisation, treatment delay, psychosocial wellbeing

For Peer Review

Introduction

Having a baby born with a cleft lip and/or palate (CL/P) can have both social and psychological impacts for parents (Nelson, Glenney et al., 2012). While generally not in the range of clinical distress, research has indicated that parents of children with CL/P may be more likely than the general population to experience parenting stress (Boztepe et al., 2020; Stock et al., 2020), as well as symptoms of post-traumatic stress (Despars et al., 2011) and postpartum depression (Grollemund et al., 2020). Further, parenting a child with CL/P can affect overall familial quality of life (QoL; Antunes et al., 2014; Macho et al., 2017; Emeka et al., 2017). The first year of the child's life, particularly around the time of primary surgery, can be an especially demanding and stressful time for new parents (Sischo et al., 2016; de Cuyper et al., 2019; Nur Yilmaz et al., 2019). In the lead up to primary surgery, parents must cope with conflicting emotions, such as the desire to help their child and improve their condition, but also the instinct to protect them from any distress caused by treatment (Nelson, Caress et al., 2012). Not only is the consideration of parental wellbeing important in itself, the welfare of the parent(s) also influences of the welfare of the child. Parental psychological distress is considered a key risk factor for a range of negative child outcomes, including medical adherence, psychological wellbeing, and social and cognitive development (Sanger et al., 2015).

In the United Kingdom (UK), specialist CL/P care is available to all families free of charge at the point of access. Following an antenatal or postnatal diagnosis of CL/P, families are typically contacted within 24 hours by a Clinical Nurse Specialist (CNS). The CNS will visit the family in the hospital or at home to provide initial information about the condition and its treatment, to offer emotional support, and answer any questions. Generic community care from midwives and health visitors, responsible for assessing child and maternal wellbeing and health, is also offered to all new families at this time. In addition, the Cleft Lip and Palate

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Association (CLAPA) is a leading UK charity dedicated to supporting those affected by CL/P that offers a range of resources for new parents.

Typically, the family will be invited to a multidisciplinary consultation during the early months, where they meet key members of the CL/P team to talk through their child’s treatment plan. The standard treatment pathway indicates that primary surgery should be performed within the child’s first year of life (National Health Service (NHS) England, 2013). These guidelines stipulate that a cleft lip should be repaired before 6 months of age, while palate repair should be performed prior to 13 months of age (NHS England, 2013). There is now considerable evidence to demonstrate the importance of this standardised pathway for optimal child and parent outcomes (e.g. Ness et al., 2015).

The Covid-19 pandemic has had a significant impact on healthcare provision and utilisation worldwide (Moynihan et al., 2021). To date, the UK has announced three national ‘lockdowns’/stay-at-home orders (26 March 2020, 5 November 2020, and 6 January 2021). There have also been numerous local lockdowns, affecting only specific geographical areas. For UK CL/P care, the pandemic has meant a disruption to routine clinic and outreach appointments, as well as significant delays to primary surgery. Initially, all non-essential surgeries in the UK were postponed. Later, guidelines were published by the Royal College of Surgeons on the prioritisation of surgery during the Covid-19 pandemic (Federation of Surgical Speciality Associations, 2020). Four priority levels of surgery were set. At the time of the interviews, cleft palate repair was categorised as ‘Priority 3’, to avoid breaching the national standard of 13 months of age. Cleft lip repair, along with most other CL/P surgeries, was categorised as ‘Priority 4’ (i.e. the lowest priority), which can wait indefinitely).

This shift in care poses important questions about the impact of the Covid-19 pandemic on the new generation of children born with CL/P, as well as their parents. The present study

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Commented [NS2R2]: We have now included further information to clarify how ‘Category 3’ and ‘Category 4’ were defined.

At the time of interview, no national data were available on the degree of the delay. This data is now beginning to be captured and we have included a reference to a poster that was presented at the recent UK craniofacial conference later in the manuscript.

It is difficult to comment on the situation of the individual participants. Cleft teams across the UK vary slightly in their protocols (e.g. one team may normally perform cleft lip repair at 3 months, while another may perform the same repair at 6 months. Some teams prioritise bilateral cleft lip over unilateral cleft lip, and ‘complete’ palate over submucous palate, for example. We could also not be sure that parent report would be accurate. Further, most parents were still waiting for a date for surgery, so the delay at the time of interview was indefinite. We agree it’s important to include something about this and have included additional text in the manuscript to address this point as best we can.

aimed to assess the impact of the Covid-19 pandemic on new parents of children born with CL/P in the UK prior to or during the initial 'wave' in the first half of 2020.

Method

Ethical considerations

Ethical approval was granted by the Faculty Research Ethics Committee at (*institution*), (*city*), UK. The ethical guidelines of the British Psychological Society were also followed at all times.

Design

This study employed an inductive, qualitative approach. Individual, one-to-one interviews were conducted with parents of children with CL/P born prior to or during the first 'wave' of the pandemic, between January and June 2020. A semi-structured interview schedule was created, based on prior literature focused on the wellbeing of parents following a diagnosis of CL/P in their child (e.g. Nelson, Glenny et al., 2012) and clinical experience. The interview schedule included questions related to: experiences of pregnancy and birth; experiences of diagnosis; experiences of healthcare from both non-specialist and CL/P specialist care providers; parental emotional wellbeing; experiences of support services; treatment-related experiences; and overall reflections, plans, hopes, and concerns for the future. See Table 1 for further information regarding the interview schedule. Demographic information was also collected.

Questions were open-ended and the interviewers were able to prompt interviewees to provide more details where appropriate. Interviews were conducted between September 2020 and October 2020 by the first and second authors, who are both trained in qualitative methods. Interviews were carried out either on Microsoft Teams ($n=2$) or over the telephone ($n=10$), and were between 27 and 56 minutes in length (53 minutes on average).

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Procedure

Participants were self-selecting and responded to advertisements published on relevant websites and social media. Potential participants who expressed an interest in the study were screened for eligibility (i.e., UK-based, over the age of 18 years, good level of written and spoken English, and had a child born with CL/P between January and June 2020) and sent a Participant Information Sheet, containing further details about what participation in the study would entail and key ethical information, such as confidentiality and their right to withdraw. Participants who agreed to take part were asked to provide verbal consent prior to the interview. The interviews were audio recorded and transcribed verbatim by the first, second, and third authors. All eligible participants who volunteered during the recruitment period (August to October 2020) were interviewed and data were collected until no new information was forthcoming.

As part of the wider study, health professionals and members of CLAPA staff were also interviewed about their experiences of providing care for new families during the first ‘wave’ of the pandemic. These findings are presented in a separate paper (*under review*).

Participants

Fourteen parents, from a total of 10 families participated. Participant demographics and sample representativeness are described in Table 2.

Analysis

Data were analysed using inductive thematic analysis (TA; Braun and Clarke, 2006). As per Braun and Clarke’s six prescribed stages of TA, the first author became familiar with the data (1) and generated initial codes (2). Codes were then collated into themes (3). The first, second, and third authors reviewed the themes and discussed them with the senior author (4),

which was aided by the creation of a thematic map. Last, final themes were defined and named (5), and the manuscript was produced (6).

Results

Thematic analysis identified three themes, each with a number of sub-themes (see Figure 1). Each theme and subtheme are presented below alongside exemplar quotes. Shortened quotations are indicated by [...] and additions for the purpose of clarity are indicated by (...). Participants have been given pseudonyms to protect anonymity.

For transparency, guidelines around quantifying language have been adhered to (Hill et al., 2005). “All” refers to all or all but one, “most” refers to more than half, and “some” refers to less than half but more than two.

Theme 1: Changes to Healthcare: The Impact of Restrictions and Reduced Contact

Perinatal Care

Like many parents of infants born in lockdown, participants in the current study discussed the impact of the Covid-19 pandemic on their perinatal care. Participants who had experience of having had a baby prior to the pandemic were able to identify some key changes in perinatal care in response to the threat of the Covid-19 pandemic. For example, participants talked about the impact of visiting restrictions, for example: *“Last time (I had a baby) there were visiting hours on the ward, whereas this time there was nothing. [...]. It was difficult, especially after having a (Caesarean section) because I was trying to do everything on my own” (Sandra)*. Participants also discussed feeling limited in what they were allowed to do while in hospital: *“I couldn’t leave my room, to go and make myself a cup of tea. So [...] if I wanted anything at all [...] I just had to ring the bell and wait.” (Monica)*

Commented [JA3]: Think this is an example of a quote identified by reviewer that is stating there were visitor restrictions, which is widely known and stated in the preceding sentence...did Sandra say anything about how she felt about this or some personal interpretation that you can add? Please review transcripts to see if the impact of descriptions throughout can be included.

Commented [NS4R4]: We have reviewed all quotes and extended them where possible, and/or provided additional commentary.

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Most participants felt the staff at the hospital had less time to offer support to mothers following the birth of their baby. For example, Edward said “(My partner) did find it to be a pretty bad experience for the couple of days she was in (the hospital). [The midwives] were so rushed... She missed a lot of her medication as well because she didn’t happen to be in bed. She was off trying to breastfeed (our son), little did she know when she came back that they’d been offering medication, so she hadn’t had pain medication for 12 hours or something. I would say that Covid pretty dramatically affected that process.” Similarly, Steven noted, “I was getting phone calls from my wife because the nurse had left her and she was in pain.” Participants also felt their time in hospital had been very brief due to reduced turnaround times. Some felt they hadn’t had enough time to recover from the birth: “They let us out quite quickly. I only stayed in one night, but it was difficult, especially after having a C-section, because I was trying to do everything on my own.” (Monica)

Once participants had left hospital and returned home, many felt that essential community care, such as support from health visitors, had been adversely affected, which had a negative impact on parents’ postnatal experiences: “My health visitor (and I), we weren’t ever in (physical) contact with each other. She had to stand and wait outside... All of these times she’s been coming to my house, but she’s actually not laid eyes on (my son).” (Anna)

Care from the Specialist Cleft Teams

Parents of infants born with CL/P would typically receive in-person support from their designated CNS, particularly during the first few months of the baby’s life. Some participants reported having seen their CNS in person, wearing personal protective equipment. Lucy commented, “I have now seen the cleft nurse [...]. She came to visit which was pretty great.”

Most participants, however, reported reduced (or no) in-person support from the CL/P team, particularly with regard to feeding, as Mike stated: “Before this lockdown [...] (the CNS) said

Commented [JA5]: Implied that this is difficult for Anna – anything she said to that effect?

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10 *'if you have trouble feeding, one of the nurses can come out and help'. But obviously when*
11 *lockdown happened none of that support was available to us [...] which obviously made it a*
12 *little bit more difficult because, if [...] a nurse could [have] come out and shown us what to*
13 *do, it would have been a lot easier."* Similarly, Mary commented, *"I think (my son) would*
14 *have been assessed a bit more by the cleft nurses had they been able to do home visits... I*
15 *personally would have been happier with a lot more physical support with feeding. Especially*
16 *in the early weeks... because it is really different from feeding a child without a cleft."*

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23 Most participants reported positive experiences of the support they had received from the
24 CL/P teams, in spite of the pandemic. As Anna expressed, *"We certainly felt supported. I*
25 *can't thank (the CNS) enough. She's always been at the end of the phone and she's always*
26 *communicated things [...]. She didn't always know what was happening either, and she was*
27 *completely honest about that."* Sandra also stated, *"I really don't feel like I have been*
28 *abandoned or anything [...]. I definitely feel (the CL/P team) has been (on top of it)."*

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33 However, some participants reported less positive experiences, particularly in regard to
34 communication. Edward reported, *"I don't think we've had any further contact from the cleft*
35 *team for months."* Similarly, Monica said, *"(The nurse) knows about the anxiety I'm having,*
36 *but I've still had to ask about everything [...]. The communication has not been very good*
37 *[...]. They haven't bothered to get in touch at all [...] and see if I'm doing OK."* Some
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42 participants felt they had to make a considered effort to stay in touch themselves: *"(The CL/P*
43 *team has) always been good at communicating when I've asked a question, but not*
44 *necessarily coming forward and telling me things."* (Lindsey)

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48 Nonetheless, participants recognised and empathised with the challenges faced by the CL/P
49 team during the pandemic: *"(The CL/P team have) got to deal with a lot. I imagine that*
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Experiences of Virtual Consultations

Some participants talked about remote consultations positively, expressing gratitude that the support was available at all: Tommy said, *“It’s better than being left alone in the dark and not having anyone to talk to about it,”* and Lindsey stated, *“I was grateful that the appointment could still go ahead.”* Participants also reported receiving remote support from community-based organisations, such as CLAPA, which appeared to be a source of information and advice: *“I browsed the CLAPA website. It gives a lot of information, it’s packed with knowledge in terms of what to expect and [...] there is also a family community where you can reach out.”* (Michelle) Jackie similarly reported, *“We have been using the [...] online coffee chats that (CLAPA) do [...]. I’ve listened to the diagnosis ones and the feeding ones and the surgery one. [...] When I’ve listened to other people’s experiences it helps me [...] think ‘OK, yeah, they’ve dealt with it, that’s what happens’.”* Participants also referenced using social networking sites to access information and support, as Edward noted, *“The only information we’ve been able to glean is from other parents in a Facebook group.”*

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Everyone is in the same boat trying to help each other out. But it really heightened the anxiety and (made me) compare... It's all just more worrying." (Lucy)

Theme 2: Family Functioning During the Pandemic

Parental Anxiety

Unsurprisingly, parents reported experiencing emotional challenges during the first few months of their baby's lives, as Steve reported: *"I remember one morning, I'd been up most of the night with the baby [...]. My wife is crying, baby's crying [...]. I just stepped out the room. I remember I sat with my head in my hands on the stairs, just thinking 'it's not meant to be like this'."* It is not uncommon for new parents, particularly those whose children have additional health needs, to experience emotional challenges. However, participants' accounts suggested that these struggles were heightened by the pandemic and reduced access to support: *"(Cleft lip and palate) is quite a big thing to be dealing with for any parent [...] and then, add onto that as well, that we have a global pandemic."* (Monica)

The Experiences of Fathers

During the first 'wave' of the pandemic, a one-parent policy was implemented in most hospitals. This resulted in many fathers having limited or no involvement in prenatal appointments, during the birth, and at in-person consultations. Mothers in this study described feeling overwhelmed at having to experience these events alone, particularly if they were first-time mothers: *"(My husband) left me at the entrance door (of the hospital). I had to carry my luggage and my big belly and everything to the first floor and I was quite emotional because I was left on my own."* (Michelle) These limitations were also impactful for the fathers themselves. Steve said, *"(During the birth) I waited for three hours in the car park [...]. That was scary."* Similarly, Mike noted, *"He is my son, I would like to be as involved as*

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10 (my partner is) [...] Normally if there were any appointments or anything I would be there
11 [...] (I just had) to go on the feedback that (my partner gave) me.”

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14 Positively, participants talked about fathers spending more time at home with the family due
15 to lockdown restrictions. This allowed fathers to offer practical help in caring for the baby,
16 and spend more quality time as a family, as Steve reported, “(I had) four months (working
17 from) home with the baby [...]. I saw her every day [...]. I will proudly say I was the first
18 person to make her smile.” Edward similarly stated, “I was working from home, so I had
19 quite a nice time where I could be there to help. There's always going to positives [...].
20 That's one of them [...]. I guess that's a silver lining.”

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27 Support from Family and Friends

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29 Due to the restrictions imposed by the ‘stay-at-home’ order, participants were unable to rely
30 on support in person from family and friends during their child’s first few months of life:
31 “(Support from family) has been limited due to the restrictions [...]. I know that they are
32 there for some emotional support, but in terms of how much they have been involved in (the
33 baby's physical) care, it has been limited because of Covid.” (Lindsey). Steve also expressed,
34 “It was very much, just me and my wife [...]. We didn't really have anyone [...] It was
35 difficult to say the least.”

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38 One parent commented that the lack of support she received in the beginning was so
39 challenging that she might have been discouraged from having more children: “(My mother
40 and mother in law were) talking (to me) [...] on the phone, but it's hard... It was really
41 difficult. If he had been my first child I'd never have had another.” (Mary)

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49 Nonetheless, some participants were able to find positives during this period of isolation: “In
50 some respects it was good, because you don't feel like everyone's dropping in all the time and
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you and can really get to know the baby, and get to know their routine and what they're like.”

(Sandra)

Theme 3: Surgical Prioritisation: Delays and Uncertainty

Changes to Surgical Protocols

All participants reported that their child's primary CL/P surgery had been delayed due to the Covid-19 pandemic. For most children, surgery had been delayed indefinitely and parents were still waiting on a surgery date. The issue of surgical prioritisation was frequently discussed. Overall, participants were understanding about the need to prioritise certain patients during the pandemic: “(The CL/P team) said that there is a priority list, so it's obviously the more urgent ones that are going to get done sooner... We didn't really question it because I totally agreed with that.” (Angelina)

However, all participants still expressed disappointment and frustration about their child's surgery being delayed, Mike said, “The negative thing [...] is the delay on getting the surgery date.” Monica also stated: “It just feels a bit like [...], why is it taking so long to start back up again? And surely there can't be that much of a backlog. It's getting a bit frustrating. Why can't they figure out a system that works better?”

In addition to the need to demonstrate a negative Covid test prior to coming into the hospital, most participants also talked about the need to self-isolate. As Angelina noted, “We have to have the whole family in quarantine for two weeks before (the surgery) and with my partner I'm not sure... I don't know how we're gonna do that, because he's a company director, he's got to run the business.” Tommy explained the difficulty of isolation requirements as well: “We (were told) to take two weeks off work (to isolate). I didn't get to see (my son) for two weeks because I went to work and stayed at my parents' [...], so it was very hard.”

Dealing with Uncertainty

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Due to delays to surgical waiting lists, many participants expressed feelings of uncertainty around their child’s primary surgery: *“We don’t know (if the surgery) will be next week or? [...] It would be good if we could know what’s what.” (Edward)* This uncertainty resulted in an increase in parental anxiety, as Lucy stated, *“It was all just unclear and unknown, and that was really scary.”* Jackie similarly said, *“It’s a bit daunting [...]. In a normal situation, take away Covid, you would be worried and anxious for (your child’s) operation. But I just keep panicking, like oh no, what if lockdown strikes again and (my daughter’s) operations are postponed or cancelled?”*

Due to an increased reliance on remote support services, participants were able to see other parents posting about surgery on social networking sites and forums, which prompted comparisons: *“I saw that (another family’s) baby who had also had a unilateral cleft had already had their repair surgery and was done. And he was born after mine.” (Monica)*

Complications of Delaying Surgery

Most participants discussed the potential longer-term impacts of delaying primary surgery. Many agreed that managing the impact of the surgery itself would also become more difficult the older the child got, as Mike stated: *“I know (my son is) going to get his surgery done at some point [...], but the further it gets pushed back the harder it will be.”* These concerns were largely due to practical challenges of conducting the operation at a later stage in their child’s development. Rachel pointed out, *“By the time they get around to doing (my daughter’s) surgery, she’ll be just learning to walk. She might fall over and (hurt herself).”* Mike was also concerned, *“If (our son) hasn’t had his surgery done, we’re not sure - would it make it any harder to wean him off the bottles? Things like that are a concern to me and (my partner).”* Michelle noted, *“At the moment (my son) puts everything in his mouth, and that will have to be kept to a minimum so it will be a tricky time. We’ll have to make sure that the*

wound is healing well.” Other participants referred to a desire to reduce the burden of treatment as soon as possible for their child: “I just don’t want it to be dragged out for (my son). I’d prefer that [...] he got it all done.” (Tommy)

Finally, participants expressed concerns about the impact of surgical delay on their child’s longer-term speech development. Lindsey stated, “I feel like (my son) is trying to make more sounds now, trying to do more from a speech point of view, but he can’t. I feel a little bit anxious about that, wanting him to get surgery sooner rather than later for that reason.”

Tommy similarly was concerned, “It is quite sad to think that (delaying surgery) could affect (my son’s) speech [...]. The biggest concern I have is I hope it doesn’t affect (my son) long term.”

Generating Positive Meaning

In spite of the uncertainty surrounding surgery and participants’ concerns about the impact of treatment delays, most participants highlighted positives of being able to spend more time with their baby prior to primary surgery with appreciation of their child’s cleft, as Steve noted, “(My daughter’s) smile is the best thing in the world, it really is. Just to have a cleft smile for a (while longer), that’s nice.” Anna similarly expressed, “I’m just so thankful I’ve got [...] a good seven months of (my son) on my phone camera roll, of just him and his smile on his face the way it was.”

Discussion

The Covid-19 pandemic has disrupted healthcare around the world, and has impacted CL/P care in the UK significantly. The present study aimed to assess the impact of the Covid-19 pandemic on new parents of children born with CL/P in the UK prior to or during the initial ‘wave’ in the first half of 2020. The findings provide insight into the range of heightened and

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additional challenges that parents experienced, from pregnancy through to primary surgery. Findings are summarised below and key learning points are highlighted.

The Emotional Impact of the Covid-19 Pandemic on Parents of Infants with CL/P

The birth of a baby with CL/P is an emotional and challenging time for most parents, as they navigate the implications of the condition and its treatment (Nelson, Glenny et al., 2012). The findings of this study demonstrate the potential heightened emotional impact brought about by the Covid-19 pandemic. Uncertainty regarding their child’s primary surgery, in addition to reduced contact with the CL/P team and other health professionals appeared to increase anxiety and emotional distress among participants. Further, due to the ‘stay-at-home’ order, participants found themselves separated from friends and family, as well as unable to access other sources of support in person, such as ‘parent and baby’ groups, which would normally offer crucial support during pregnancy and after birth. A lack of social support that would typically be more readily available in person is not only associated with higher rates of maternal depression (Sank et al., 2003) and stress (Boztepe et al., 2020), but is also instrumental for coping (Sischo et al., 2017) and increasing resilience in parents (Yu et al., 2021).

Between April and June 2020, a large-scale survey of the general UK population of new parents identified a significant rise in mental health concerns among new parents compared to before the pandemic began (Saunders and Hogg, 2020). Almost 9 in 10 (87%) respondents reported feeling more anxious as a result of the Covid-19 pandemic and lockdown. Two thirds of new parents (69%) felt their ability to cope with having a new baby had been impacted by the pandemic, with many reporting intense feelings of loneliness and isolation. Just over a quarter (26%) had concerns about bonding with their baby, with a similar number also reporting relationship challenges with their partner. Concurrently, access to antenatal and

perinatal healthcare was reduced, with only 11 percent having seen a health visitor in person. Fewer than 3 in 10 (32%) respondents felt confident they could find help for their mental health concerns. These findings may be particularly challenging for parents of children born with a health condition, such as CL/P, where the child has additional medical needs, and the psychological impact on parents is already known to be greater than in the general population (Stock et al., 2020).

As in the present study, the UK report (Saunders and Hogg, 2020) identified a unique impact on fathers. Involvement in routine appointments, antenatal classes, and the birth, as well as the confidence gained through early interaction helps fathers to bond with their baby and build stronger a relationship with their partner (Meier and Avillaneda, 2015). The report by Saunders and Hogg (2020) highlighted how the exclusion of fathers during the pandemic has caused additional stress and worry, resulting in expectant mothers feeling unsupported and fathers feeling unvalued. Within the CL/P literature and prior to the Covid-19 pandemic, concerns around the exclusion of fathers have been specifically highlighted. Studies have suggested that having a baby with additional needs makes the gap in support for fathers more evident (Nidey et al., 2015; Stock and Rumsey, 2015; Zeytinoglu et al., 2016), with fathers of children with CL/P reporting less satisfaction with healthcare communication and inclusion (Costa et al., 2020). Further, in a study by Stock and Rumsey (2015), fathers believed strongly that their key role in the CL/P process was to support their partner and new-born baby. As the current study demonstrates, the pandemic reduced fathers' opportunities to support their partner and baby in the ways they would have liked by excluding fathers from many key medical appointments. Nonetheless, participants in the current study were able to identify some positives associated with fathers being at home more after the birth. These included more opportunities to bond with the baby, a chance for fathers to offer practical

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support in caring for their baby, and more time spent as a family; findings that were also identified in the general UK population (Saunders and Hogg, 2020).

Receiving Support Remotely

While routine CL/P care is often rated highly by new parents (Costa et al., 2020), evidence of dissatisfaction with healthcare was noted in the current study. Specifically, in-person appointments were not possible, resulting in reduced overall contact with the CL/P team and other health professionals, and CL/P consultations being carried out remotely. Although some participants were grateful to have the option of remote contact, many highlighted a lack of support for feeding difficulties in particular, an area known to impact upon parental wellbeing, parent-infant bonding, and child health (Costa et al., 2019; Madhoun et al, 2020). Extensive previous research has demonstrated the importance of support from healthcare professionals for parents following a diagnosis of CL/P. One large study based on UK cohort data found that satisfaction with healthcare was protective against psychological distress in both mothers and fathers of infants with CL/P (Stock et al., 2020). Two other quantitative studies also highlighted an association between reduced access to care and poorer parent and child outcomes (Bennett et al., 2018), and between parents’ relationship with their CL/P team and healthcare satisfaction (Byrnes et al., 2003). Qualitatively, Searle and colleagues (2018) highlighted the key role of the CNS specifically as a highly valued and trusted contact and as a mediating link between the family and the rest of the CL/P team. Conversely, and as illustrated by the current study, the absence of this support and the remote nature of contact with health professionals can have a detrimental effect on wellbeing. A related paper detailing interviews with UK health professionals during the first ‘wave’ of the pandemic demonstrated this was also a source of concern for CL/P nurses, who responded by conducting more telephone calls than usual to ensure they kept in contact with families as much as possible, particularly with those considered to be at higher risk (*under review*).

Many participants in this study had accessed online support, such as the peer support services offered by CLAPA and other online forums/social networking sites. Before the pandemic, CLAPA offered regular opportunities for face-to-face support, such as regional 'family days' and fundraising events, which were adapted to online support during the first lockdown. Although online support can give parents opportunities to share their experiences, connect with others, give and receive emotional support and information, and obtain quick responses to queries in a semi-anonymous environment (Stock et al., 2018), studies have also shown that online information can be of variable quality (Antonarakis and Kiliaridis, 2009). In addition, not all parents benefit from participating in online forums (Stock et al., 2018), as also shown in the current study.

Surgical Prioritisation

The first year of the child's life, particularly around the time of surgery, can be a particularly demanding and stressful time for new families (Nelson, Caress et al., 2012; Sischo et al., 2016). The standardised treatment pathway and the specialist support offered by the CL/P teams normally act as a source of reassurance (Nelson, Glenny et al., 2012). As a result of the Covid-19 pandemic, treatment pathways were disrupted and specialist support was reduced and/or delivered remotely. Initially, all but emergency surgery, including CL/P surgery, was paused altogether. Once surgery in the UK restarted, most CL/P surgeries were considered to be relatively low priority, resulting in less resource allocation and prolonged waiting lists. One ongoing observational study in the UK recently reported an increase in the

time between birth and cleft lip surgery of 93.4 days, and an increase in the time between birth and cleft palate repair of 67.5 days, from March 2020 to April 2021 (Brierley et al., 2021). These data related to 651 records from five UK CL/P teams, and is therefore not representative of the UK as a whole, but does provide an indication of the emerging picture.

Participants in this study were largely understanding of the need to prioritise surgery and

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make the best use of restricted NHS resources. However, this did not fully mitigate the frustration and anxiety felt by parents in relation to surgical delays. Participants expressed concerns for their child’s immediate and longer-term development, as well as a wish for clarity on the situation to support their own mental health. Potential complications of performing surgery later were raised (such as the child’s increasing mobility), alongside the practical challenges of the need to self-isolate and test for Covid-19 exposure prior to entering the hospital. In addition, participants had become aware through social networking sites of other hospitals in the UK that had been able to ‘catch up’ on their waiting lists more quickly than others. This had exacerbated parents’ emotional distress and reduced their faith in their local CL/P team, as well as highlighting potential inequities in care across UK regions.

Implications for Research and Clinical Practice

At the time this study was conducted, and based on annual incidence rates (CRANE, 2020), an estimated 500 infants with CL/P would have been born in the UK immediately before or during the first wave of the pandemic. At the time of writing, one year later, a further 1,000 infants will have been born with CL/P in the UK. Furthermore, an additional number of children born with CL/P in the latter part of 2019 may also have experienced surgical delays. The disruption to CL/P care in the UK has resulted in an accumulation of children requiring primary surgery. While the extent of this backlog will vary between CL/P teams, due to differing levels of resources and prevalence rates in different regions, the potential current and ongoing impact on the parents, the wider family, and the affected child should not be underestimated.

At the time of interview, cleft palate repair was prioritised over cleft lip repair, largely due to the evidence to show speech development may be affected if palate repair is not performed

before 13 months of age (Watson et al., 2001). Yet, cleft lip repair is also a time-sensitive operation, with existing evidence to suggest that delays could lead to disrupted parent-infant interactions, increased parental anxiety, and prolonged isolation from social interactions (Hammond, 2021), some of which has also been indicated by the present study. Calls have been made to reassess the surgical priorities for CL/P, by not only taking medical needs into account, but also the developmental stage of the child and the potential psychological impacts on both the child and the parent (Hammond et al., 2021; *under review*).

Based on the findings of the current study, previous CL/P research, and the wider literature, health professionals should be aware of the increased potential psychological impact of CL/P during the pandemic, particularly in the face of unpredictable healthcare delivery. Parents may appreciate in-person visits and consultations wherever it is possible and safe to do so, to receive emotional support, practical support with feeding, and to develop a closer relationship with the CL/P team. Communication with parents should be candid with regard to how care has been affected, and should take regional variations in current care into account. Check-ins with families should also be carried out as frequently as possible, and referrals to Clinical Psychologists within the CL/P team should be considered, particularly for families considered to be at higher risk and for those experiencing heightened levels of distress. Regular psychological screening may help to identify those who need support the most when resources are restricted. As always, but potentially more so during the pandemic when online information is more heavily relied upon, it is important to provide parents with sources of high-quality information and for trained staff to moderate online forums where possible. Equally, health professionals could raise parents' awareness of organisations which offer various types of remote support (such as live online events and opportunities to connect with other parents by telephone), as well as available educational videos, leaflets, and books to suit parents' needs if/when they choose to access it.

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Finally, many parents in this study were able to identify positive aspects of their difficult situation. Optimism and problem-solving have been shown to be effective coping strategies among parents of children with CL/P, particularly when faced with uncertainty (Fonseca et al., 2015; Sischo et al., 2016; Stock et al., 2020). Future research could investigate the potential impact of interventions aimed at increasing these coping strategies among parents of infants born with CL/P.

Methodological Considerations

The current study is novel in its aim to investigate the impact of the Covid-19 pandemic on UK CL/P care from the perspective of parents of babies born between January and June 2020. Nonetheless, the pandemic presents an ever-changing situation, and in the last year two more national ‘stay-at-home’ orders have been instigated, in addition to numerous local lockdowns. Research should continue to investigate the impact of the pandemic on CL/P care on an ongoing basis, so that any negative consequences can be mitigated, and the longer-term implications better understood.

Other limitations of this study should be acknowledged. First, and although four fathers did take part, mothers were predominantly represented in the current sample. The limited representation of fathers in CL/P research is not uncommon and scholars have previously advocated for increasing fathers’ participation (Stock and Rumsey, 2015; Zeytinoglu et al., 2016). Second, all participants in the current sample were White. As ever, the omission of the experiences of diverse parent groups is problematic, both in terms of the relevance of the implications of the research, and in relation to understanding the needs of underserved populations (Crerand et al., 2015). This is particularly true in the case of this study, since a greater impact of the pandemic on parental wellbeing was identified in the general population, with regard to ethnicity and level of income (Saunders and Hogg, 2020). It is

imperative that future research continues to make efforts to recruit diverse samples and better understand the experiences of minority families affected by CL/P. However, the current sample was fairly representative of the UK general population overall (Office for National Statistics, 2018).

Third, none of the parents in this study reported their child to have a known syndrome. Previous research has indicated that families effected by syndromic cases of CL/P were more greatly impacted, for example with regards to a greater reduction in QoL (Feragen and Stock, 2014; de Cuyper et al., 2019). It is therefore possible that families of children born with syndromic CL/P during the Covid-19 pandemic experienced additional challenges which were not represented in this study. With regards to other demographic information about the participant's children, there was a greater representation of boys. Further, when comparing to national CL/P data (CRANE, 2020), there was also an over-representation of UCLP and CL (bilateral or unilateral), and an under-representation of CP. These are all factors to be carefully considered when gauging the representativeness of this sample.

Finally, the 14 participants in the current sample were representative of six out of 11 CL/P regional networks. This study identified a divergent impact of the Covid-19 pandemic across different regions, in part due to local lockdowns but also related to differing resourcing across CL/P teams within the UK. Future research should take these potential differences in service delivery into account.

In conclusion, this study identified a range of heightened and additional psychological and social impacts for parents of infants who were born between January and June 2020 with CL/P just before and during the first 'wave' of the Covid-19 pandemic in the UK. Parents also reported helpful coping skills, using social support networks, and generating positive meaning during challenging experiences. As the pandemic continues, the close monitoring of

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families affected by CL/P remains imperative, particularly for those at risk of emotional distress.

For Peer Review

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For Peer Review

Figure 1. Thematic Map

