

Perspectives of parents with lived experience of cytomegalovirus infection, on universal newborn screening for congenital cytomegalovirus (cCMV) in Canada: a patient-led qualitative study

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ABSTRACT

Objective To understand parental perspectives regarding universal newborn screening (UNS) for congenital cytomegalovirus (cCMV) in Canada.

Design A qualitative, patient-led study using the Patient and Community Engagement Research approach consisting of online focus groups and in-depth individual interviews to understand parental preferences regarding UNS for cCMV. Data were analysed iteratively using inductive thematic analysis and narrative story analysis.

Setting Canada-wide study conducted via video conference from October to December 2023.

Patients 12 participants from five Canadian provinces who self-identified as 18 years of age or older and as having parental lived experience with cytomegalovirus (CMV) or cCMV participated in the study.

Results We identified three themes: (1) attitudes about UNS for cCMV, including participants' unanimous support for UNS and confirmation that parental anxiety is not a deterrent for screening, (2) cCMV diagnosis, including the importance of coupling cCMV diagnosis with access to treatment and medical support and (3) awareness of cCMV, where participants shared their frustration about the lack of public and pregnant people's awareness of cCMV.

Conclusions Parental anxiety is not a deterrent for UNS for cCMV. Children with cCMV and their families deserve every opportunity to attain their best possible outcomes. UNS offers children with cCMV access to early intervention if they need it, and also helps to raise awareness and education to prevent future CMV infections.

INTRODUCTION

Cytomegalovirus (CMV) is a common virus that rarely causes concern in healthy individuals.¹ However, when a baby contracts CMV in utero, known as congenital CMV (cCMV), mild to severe life-long health and developmental consequences may result for the infant, such as hearing loss, visual impairments, seizures, developmental or motor delay, spina bifida and death.² cCMV is the

WHAT IS ALREADY KNOWN ON THIS TOPIC

- ⇒ Congenital cytomegalovirus (cCMV) is the most common non-genetic and preventable cause of birth defects and hearing loss in infants.
- ⇒ The majority of Canadian provinces do not include cCMV as part of their universal newborn screening (UNS) programmes.

WHAT THIS STUDY ADDS

- ⇒ Immense parental support for UNS programmes for cCMV to be implemented nationally as parental anxiety is not a deterrent to newborn screening, parents want the information to be able to make decisions for their family.
- ⇒ Caregivers value early diagnosis as it leads to quick access to medical follow-up, supports and resources for the best possible outcomes for children.
- ⇒ UNS for cCMV may also contribute to raising awareness for cCMV among the general public and pregnant individuals which can lead to prevention of cCMV.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

- ⇒ UNS for cCMV should be implemented nationally in Canada.
- ⇒ Raising awareness about cytomegalovirus (CMV)/cCMV should be a Canada-wide priority as every pregnant person should know what CMV/cCMV is and how to avoid contracting the virus during pregnancy.

most common non-genetic and preventable cause of birth defects and hearing loss in infants.^{2,3} It is estimated that 1 in 200 unborn children are infected with CMV during pregnancy, and 20% of those infants may go on to develop a permanent disability.^{2,3} Transmission of CMV occurs through sharing of body fluids.² Many expectant mothers who

are CMV positive, will not pass the virus to their unborn child, and even when they do, there is an approximate 75%–80% chance that there will be no consequences for the child.³ Given this reality, many healthcare providers are hesitant to discuss cCMV with expectant mothers or recommend behavioural changes, such as not kissing their toddler on the lips, as these measures may negatively impact the mother/child bonding and cause undue stress and anxiety.⁴ Early diagnosis of cCMV is necessary to obtain antiviral medications, detect health consequences and establish a variety of early supports.³

Universal newborn screening (UNS) allows all infants to be screened for various conditions that may not be evident at birth,⁵ the conditions that are screened for vary by province in Canada.⁶ The diagnostic tests typically used for UNS for cCMV are saliva, urine or dried blood spots which are tested using PCR and results are usually available within days of screening.⁷ We found no published studies on Canadian parental perspectives of UNS for cCMV. Studies from the USA,^{8–10} Italy¹¹ and Finland¹² highlight concerns associated with UNS for cCMV,¹³ including families experiencing distressing feelings after a cCMV diagnosis, doctors not having enough information about cCMV to provide in-depth information to families, adverse effects of antiviral treatments administered unnecessarily to babies, and additional burden on the healthcare system and families from unnecessary doctor visits for newborns who test positive but may not develop health concerns. Despite these findings, parents supported that cCMV newborn screening is beneficial for early detection, and subsequently, the protection of children's health and development.^{8 11 13–16} The aim of this study is to explore parental perspectives regarding UNS for cCMV in Canada.

METHODS

We used the Patient and Community Engagement Research (PaCER) approach¹⁷ to obtain rich descriptions of participants' thoughts, perspectives and experiences. The PaCER approach is patient-led and consists of three distinct stages, SET, COLLECT and REFLECT.¹⁷ SET engages other patients as partners in discussion groups to define the scope and research question. COLLECT includes various data collection activities, such as interviews and focus groups. REFLECT is held with participants from COLLECT to review study findings and ensure accuracy.

Phase 1: SET

Five patient partners with cCMV lived experience attended a 2-hour SET discussion group in May 2023 to define the study scope and research question. Key findings from SET included support for the study direction and research question, the expressed need for increased general awareness about CMV, and the position that UNS is important to diagnose and treat cCMV as early as

possible. All research team members attended the SET discussion group; RC and BT cofacilitated.

Phase 2: COLLECT

Recruitment

We strove to recruit a diverse group of parents and expectant parents from across Canada, with varying backgrounds and cCMV experiences. Inclusion criteria included individuals who were 18 years of age or older, self-identified as having parental lived experience with CMV or cCMV, and lived in Canada. Purposive sampling included advertising via social media, CMV Canada, Alberta Health Services Patient and Family Advisory Network, Alberta Strategy for Patient Oriented Research (SPOR) SUPPORT Unit and other SPOR SUPPORT Unit patient networks.

Data collection

We conducted 90 min online focus groups and 30–60 min interviews in October 2023. Our questions were tested with patient partners prior to data collection and related to participants' lived experience with cCMV, including their cCMV diagnosis journey, reflections on UNS and their insights on how these could be improved for future families. The focus groups and interviews were digitally recorded and transcribed. All participants were given a participant key to ensure anonymity. Consent forms were signed by each participant, and they were reminded of the voluntary nature of this study and their right to withdraw at any time. One team member, II, was responsible for all communication with participants.

We held two focus groups, SP and LB facilitated the first focus group and II and BT the second one. We offered individual interviews for those who were unable to attend focus groups, or who preferred an individual setting. Two team members were present for each interview; one facilitated, while the other took notes.

Data analysis

Focus group data were analysed thematically to identify and analyse patterns of meaning from the data and shed light on ideas and concepts that arose.¹⁸ For this study, we defined a theme as a specific pattern of explicit and/or implicit meaning evident in the raw data. The team reviewed the transcripts and individually created codes. We then met to review and consolidate the codes into a codebook. Team members used the codebook to code independently before meeting to establish consensus. Once coding was complete, themes and subthemes were identified and defined.

We used a narrative analysis to analyse accounts shared by participants under the assumption that people organise their daily experiences and interpretations of reality in the form of narratives that give shape to their lives and identities.¹⁹ Narrative data collected during interviews were examined for setting, timeline, characters, plot and positioning.¹⁹ As a team, we reviewed notes, transcripts and recordings from the interviews and captured two or

three main narratives from each interview using quotes from the transcript to substantiate the themes. Narrative analysis added additional context to the thematic analysis.

Phase 3: REFLECT

We invited COLLECT participants to attend REFLECT sessions in December 2023 to review the study findings and provide feedback on our reflections and recommendations. Two participants attended a 90 min focus group, and we held five individual interviews.

Patient and public involvement

This study was conducted by patient research partners; two had lived experience with cCMV. During our SET stage, we worked with five additional parents with cCMV experience to codesign the study. Results were analysed by patient research partners and presented back to participants during the last phase, providing participants an opportunity to confirm themes and provide additional insights prior to finalising results.

RESULTS

12 participants with parental lived experience with CMV/ cCMV shared their unique and diverse experiences from five Canadian provinces (see [table 1](#)).

We identified three themes from the data: (1) attitudes about UNS for cCMV, (2) cCMV diagnosis and (3) awareness of cCMV. These themes and their subthemes are presented in the following sections.

Attitudes about UNS for cCMV

This theme captured participants' thoughts and opinions regarding UNS for cCMV. It included subthemes of 'support for universal cCMV newborn screening' and 'anxiety is not a deterrent' that validate the immense support for the implementation of UNS for cCMV (see [table 2](#)). Our participants shared their concerns that not offering screening for cCMV could be considered unethical in that it unnecessarily denies families and caregivers important medical information about their baby and the opportunity for early intervention and treatment. Further, participants shared their experiences that protocols are in place for healthcare providers to follow when a cCMV diagnosis is confirmed to prevent overtreatment.

cCMV diagnosis

This theme describes participants' experiences with their child's diagnosis of cCMV. Participants reflected on pregnancy experiences and their pathways to diagnosis including the impact of screening, medical processes, communications with medical professionals, variances in health and development impacts, emotional impacts of the diagnosis, and supports and resources needed post-diagnosis. Participants described a broad range of experiences that resulted in three subthemes: 'quick access to medical follow-up postdiagnosis', 'supports and resources' and 'medical professional communication' (see [table 3](#)).

Table 1 Self-reported characteristics of study participants (n=12)

Characteristic	n (%)
Province	
Alberta	3 (23)
British Columbia	1 (8)
Manitoba	1 (8)
Ontario	6 (50)
Saskatchewan	1 (8)
Gender	
Male	4 (33)
Female	8 (67)
Age group	
25–34 years	3 (25)
35–44 years	8 (67)
55+ years	1 (8)
Area of residence	
Rural	4 (33)
Urban	8 (67)
Ethnicity	
European	9 (75)
Indigenous	1 (8)
Not stated	2 (17)
Participant type	
Pregnant person who contracted CMV	8 (67)
Partner	4 (33)
Tested positive for CMV during pregnancy	
Yes	2 (17)
No/not tested in pregnancy	10 (83)
Child's cCMV status	
Diagnosed outside of a universal newborn screening programme	7 (58)
Diagnosed through universal newborn screening programme	5 (42)
Age of child at CMV diagnosis	
In-utero	1 (8)
Newborn (0–2 weeks)	5 (42)
Early weeks (2–3 weeks)	3 (25)
Early months (2–3 months)	3 (25)
Age of child with CMV at time of survey	
Infant (1–12 months)	1 (8)
Toddler (1–3 years)	4 (33)
Preschooler (3–5 years)	4 (33)
Adolescent (10–18 years)	2 (17)
Adult (18+ years)	1 (8)

cCMV, congenital cytomegalovirus; CMV, cytomegalovirus.

One participant expressed that her infant began antiviral treatment at 3 months and does not believe it is a coincidence that her development is consistently 3 months delayed. Another participant shared that her daughter

Table 2 Subthemes for ‘attitudes about universal newborn screening for cCMV’

Subtheme	Subtheme exemplar quotes
<p>Support for cCMV screening Participants’ statements expressing immense support for universal newborn screening for CMV to be implemented nationally</p>	<p>‘I do think universal screening is tremendously, tremendously, more valuable to all the babies out there and all the parents out there that aren’t being diagnosed’. (I1-1) ‘This is a no-brainer to have universal screening across all of North America, not just Canada, because isn’t it like the largest cause of non-congenital disability in children?’. (I3-6) ‘My child passed the hearing screening, was completely healthy, no issue. So, we would have been that family that was lost. I really appreciate that we had newborn screening because otherwise I don’t know where we would be right now, what kind of position or like, where my [child] would be or if he would have any supports or if we’d still be trying to figure out things’. (I6-12)</p>
<p>Anxiety is not a deterrent Participants’ opinions expressing that families may experience stress/anxiety during the screening and diagnosis process; however, this is not a deterrent for implementing screening. Families deserve to have the information and/or access to the information, and that anxiety can be managed with support and resources. This includes participants’ assertions that families’ anxiety would be amplified in the absence of screening, receiving a late diagnosis and the resulting missed opportunities for an early intervention</p>	<p>‘No matter what the anxiety is, multiply it by a thousand if you don’t have a diagnosis and you’re looking back saying what if I could have’. (FG2-12) ‘Having that piece of mind of the diagnosis is valuable for a symptomatic kid, but even having a diagnosis for an asymptomatic kid is phenomenally valuable, because now you can watch your kid and you know what to watch for. So, the argument that parents don’t want to know and that parents aren’t responsible enough to know, and parents can’t handle the burden with this diagnosis, because they’re weak individuals or they’re not of strong personality or strong enough character to handle or to cope with this information is categorically false’. (I1-1)</p>
cCMV, congenital cytomegalovirus; CMV, cytomegalovirus.	

was diagnosed with a mild hearing loss, however, by the time they began the antiviral treatment 1 month later her daughter’s hearing loss was profound; both comments relate to the need for early diagnosis and quick access to medical treatment. Participants also related their experiences with varying levels of support (eg, psychosocial

and resources (eg, education, information) that were available and/or provided during their diagnosis journey. Some participants shared that they felt overwhelmed and alone without sufficient support and resources, whereas others expressed gratitude for the comprehensive and coordinated support and resources they received.

Table 3 Subthemes for ‘cCMV diagnosis’

Subtheme	Subtheme exemplar quotes
<p>Quick access to medical follow-up postdiagnosis Participants’ experiences that UNS prompted quick access to medical follow-up, including testing, treatment, intervention and ongoing follow-up for symptomatic and asymptomatic children</p>	<p>‘What worked well would just be how quick everything went and how supported I did feel from a medical perspective. So we had to be tested within the first month to be put on the antiviral. So, the fact that we were able to get diagnosed really early gave us the chance, especially to protect hearing and have all the tests done and then be connected with the right physicians and therapists and everything really early, I think that was really incredible’. (I6-12) ‘Within 24 hours we saw 4 different clinics and got pretty quick service there, and all of them were saying we’re just gon confirm this looks like typical cCMV, and then I think they found it within a week because they drew from the initial blood spot’. (FG1-5)</p>
<p>Supports and resources Participants expressed mixed perceptions of supports and resources available to families during their cCMV journey. Two out of twelve participants felt they received adequate support while the remainder expressed concerns regarding insufficient support (eg, psychosocial) and resources (eg, education, information) provided and/or available during their child’s diagnosis journey. This also included a need for more cCMV appropriate support and information to accompany screening and diagnosis</p>	<p>‘You know, it was here’s a diagnosis, but we don’t have anyone to tell you more about it. It was sort of what was given to us, and it was sort of we’re on our own to sort of navigate the world’. (I3-6) ‘We have so many questions as a family that knew nothing about CMV, and it’s been hard to access that support since her diagnosis’. (FG2-9).</p>
<p>Medical professional communication Experiences of participants regarding how professionals communicated the cCMV diagnosis and/or prognosis to families. This included concerns about insensitivity and lack of empathy and statements expressing the need for further professional education and training for medical professionals</p>	<p>‘And just like very like frivolous ... not thinking through these words that really took like a toll on me, telling me that I could have done something and now you’re telling it it’s not something that was helpful and, you know, just being so casual about all of these patients that he’s had and I’m talking specifically about one person right, and it’s just but being trained on the dialogue of CMV’. (FG2-12) ‘She looked at me, she was like, well, it’s normal for you to feel that way given your circumstances, and they left it, and that was that’. (FG1-4)</p>
cCMV, congenital cytomegalovirus; CMV, cytomegalovirus.	

Table 4 Subthemes for ‘awareness of cCMV’

Subtheme	Subtheme exemplar quotes
General lack of awareness about CMV Participants reported experiences relating to the general lack of education and awareness about CMV/cCMV, especially given its prevalence	‘The awareness is just not there. Like I remember when she was diagnosed, they told us that it was more common than Down syndrome, but everybody knows what Down syndrome is’. (FG2-9) ‘Nobody ever even told me that it was, it was dangerous to have any virus or cold, you know? Whereas you knew about certain things, like hand, foot, and mouth, but it’s just kind of crazy that I didn’t, I didn’t even know that CMV could be an issue’. (I6-12) ‘I think that would go a long way as far as helping people with their anxiety and manage that anxiety just to know that the odds are with them’. (FG1-4)
Lack of awareness about CMV infection during pregnancy Opinions reinforcing the need for more widespread awareness and education across systems during pregnancy and assertions that education and awareness will assist in prevention of cCMV as well as decrease anxiety for parents	‘Why isn’t this something my GP or my OB ever shared with me? How to prevent it?’. (FG1-5) ‘There’s so many things up in their office, on like herpes and warts and all these kind of things you can get while you’re pregnant, but not CMV?’. (FG1-3) ‘It was very frustrating. I had done my research. I had followed all the rules during pregnancy. I just didn’t know this was something I needed to be working to avoid. So, it was hard to get past some of those anger pieces. Why don’t we know about this? Why isn’t this common knowledge? So that was tough. And then of course, it sets in, what is CMV? I had never heard of it before’. (FG1-5)
Universal newborn screening for cCMV improves awareness Beliefs that UNS for cCMV would prompt healthcare professionals to provide more information and in turn, allow parents the opportunity to inquire about, and raise their awareness of CMV/cCMV. Participants said UNS for cCMV will help increase awareness for families with a positive diagnosis of CMV and the general population	‘I think it’s just so important that we explain to families why this particular virus should be added to newborn screening and what it means for you’. (FG2-9) ‘I think in that sense it can contribute to awareness for sure, and I think there is more conversation happening across the country now in relation to CMV, specifically being added, or whether or not it should be added to the newborn screening’. (FG2-9)

cCMV, congenital cytomegalovirus; CMV, cytomegalovirus.

Awareness of cCMV

This theme encompassed the participants’ thoughts regarding the overall lack of education and awareness around CMV/cCMV and was consistently reinforced by all participants. Three subthemes captured participant experiences: ‘general lack of awareness about CMV’, ‘lack of awareness about CMV infection during pregnancy’ and ‘UNS for cCMV improves awareness’ (see [table 4](#)). Participants expressed frustration and/or anger about not being informed of CMV/cCMV during pregnancy. Participants shared that they followed healthy pregnancy guidelines, but their providers did not inform them about cCMV, so they did not know about preventive measures that could have profoundly changed their children’s future health and development. Some participants shared feeling guilty about transmitting CMV to their fetus, and that guilt was longstanding, even while acknowledging they had not known how to protect them.

Recommendations

Participants identified three key recommendations during COLLECT, which were confirmed and refined with participants during REFLECT (see [box 1](#)). Participants expressed deep passion about the third recommendation and suggested specific campaigns for targeted audiences, including: (1) provide pamphlets to centres where expectant families visit (ie, doctor’s offices; public health units; daycares, etc), (2) present at medical/nursing schools, public health or related conferences, (3) develop digital content and infographics for medical,

childcare and/or provincial healthcare websites, (4) engage traditional media, (5) post on social media (ie, public health, ‘mommy and me’ sites, etc) and (6) inform public health officials and government ministries.

DISCUSSION

To the best of our knowledge, this is the first patient-led, Canadian, qualitative research study to explore parental preference regarding UNS for cCMV. Participants experienced various pathways to their CMV/cCMV diagnoses, however, regardless of how participants received their CMV/cCMV diagnosis, and whether their children were experiencing cCMV-related health and developmental impacts, they unanimously supported UNS.

Box 1 Recommendations

1. Universal newborn screening for cCMV should be implemented nationally in Canada.
2. Medical professionals should provide coordinated and comprehensive resources and support at the time of the CMV/cCMV diagnosis, with established protocols and processes to promote ease of access and support positive diagnosis.
3. Raising awareness about CMV/cCMV should be a Canada-wide priority. Every pregnant person should know what CMV/cCMV is and how to avoid contracting the virus during pregnancy.

CMV, cytomegalovirus; cCMV, congenital cytomegalovirus.



The support for UNS for cCMV aligns with other studies, in which most participants supported UNS and that the initial worry caused by UNS is not a reason to not screen or discuss it with parents.^{8–10 20–22} Our participants strongly expressed that anxiety should not be a deterrent for UNS. Our participants agreed and recognised that receiving a cCMV diagnosis could cause undue stress, worry or anxiety for some families whose children may not develop cCMV related health or developmental impacts; however, the benefit of avoiding worry for some parents does not outweigh the cost of lost opportunities for other children's early intervention and treatment. Our participants expressed that knowledge is power, and that knowing about a cCMV diagnosis gives families the best opportunity to support their children to become the best version of themselves.

Additionally, participants stated that quick access to treatment and early intervention is essential to prevent or lessen the development of cCMV symptoms. They also noted that unnecessarily withholding this information and associated interventions would be unethical. Although participants were unanimous in their support for UNS, some participants' support was at least partly contingent on health systems adequately supporting families throughout the cCMV diagnosis journey and beyond. It will be important to ensure that support and resources are provided and/or available, given the increased demand that UNS for cCMV will create.

Our findings call for medical professionals to be supportive, informative and strongly consider how they deliver a cCMV diagnosis to families, as the news impacts them deeply and are consistent with previous findings.^{10 13} Some participants experienced excellent medical care and support, other participants struggled with the way their health professionals communicated their cCMV diagnosis. UNS for cCMV requires increased professional education and training, as more health professionals will be supporting families through the cCMV diagnosis and follow-up journey.

Although the Society of Obstetricians and Gynaecologists of Canada's practice guidelines recommend educating pregnant people on CMV infection and hygienic measures to prevent acquisition during pregnancy,²³ our participants had significant concerns regarding the lack of cCMV awareness during pregnancy. Whether diagnosed years ago or recently, participants asserted that the ongoing lack of awareness is unacceptable and expressed frustration and/or anger about not being informed of CMV/cCMV. It was difficult to reconcile the effects of cCMV in their children and realising transmission could have been prevented. As cCMV becomes part of newborn screening programmes, media and health ministries may pay more attention and there may be increased research interest, therefore, UNS could be a catalyst for widespread awareness.

Our recommendations align with our three themes and could be implemented using the integrated care model.²⁴ According to the WHO, integrated health

services allow people to receive a continuum of health promotion, disease prevention, diagnosis, treatment, management, rehabilitation and palliative care, coordinated throughout their lives.²⁵ In relation to the integrated care model,²⁴ UNS for cCMV would potentially prevent or lessen the severity of health and developmental impacts on children with cCMV by facilitating a quick diagnosis, access to treatment and disease management, and provide for coordinated short-term and long-term supports for symptomatic and asymptomatic children.

We ensured the dependability of the study by consulting patient partners at the outset, developing a detailed protocol, documenting the process and changes, coding data individually and then collaborating to establish data consensus and member-checking the post-data analysis to validate results. Confirmability was achieved through individual reflection as well as team discussions about bias. This study included several male/paternal perspectives which is an area that not many other studies included. Our study had a small number of participants and would have benefitted from more diverse perspective additional perspectives of caregivers whose children had received a cCMV diagnosis, but who did not currently exhibit symptoms. However, qualitative research aim is to not reach generalised findings, but to gather rich descriptions of their thoughts and perspectives to further understand their lived experiences.²⁶

Children with cCMV deserve every opportunity to realise their best possible outcomes. UNS offers children with cCMV an effective and low impact opportunity for early intervention, as well as short- and/or long-term follow-up. Screening can promote comprehensive and coordinated support and resources from well-informed medical professionals, and lead to increased public awareness and education to prevent future transmission, as well as equity to better outcomes.

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Patient consent for publication Not applicable.

Ethics approval This study involves human participants. On receiving approval from University of Calgary Conjoint Health Research Ethics Board (Ethics ID number REB23-0878), the remaining two stages of the PaCER research process commenced. Participants gave informed consent to participate in the study before taking part.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement No data are available. Our ethics approval does not allow us to share raw study data.

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