

# Assessing the Burden of Congenital Cytomegalovirus Disease: Methodology to Develop a Survey Study of Patients and Caregivers in the United States

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## BACKGROUND

- In the United States, congenital cytomegalovirus infection (cCMVi) is an important cause of permanent disabilities and death.<sup>1</sup>
- Among children born with cCMVi, approximately 1 in 5 may develop permanent problems such as hearing or vision loss, lack of coordination, or developmental disabilities due to symptomatic congenital cytomegalovirus disease (cCMVd).<sup>2</sup>
- The quality-of-life impacts and economic burden of cCMVd on the patient, caregiver, and family are thought to be substantial, particularly for patients with severe impairment.<sup>3</sup>
- The types of costs that a patient with cCMVd may incur throughout his/her life may be broken down into four categories: direct medical costs, direct non-health care costs, indirect costs, and intangible costs.<sup>4</sup> [Please see the poster "A Framework for Assessing the Lifetime Economic Burden of Congenital Cytomegalovirus in the United States," presented at the 2018 CMV Public Health and Policy Conference, September 24, 2018, 4:30 p.m. to 6:00 p.m.]

## OBJECTIVE

- The objective of this work was to begin development of a cross-sectional patient and caregiver survey to collect data on the economic and humanistic burden of cCMVd, in order to describe quality of life impacts and costs for medical travel, unpaid caregiver time, education, special needs housing and lost work productivity.

## METHODS FOR DEVELOPING CONTENT OF THE PRELIMINARY QUESTIONNAIRES

- Standard survey methodology was employed for the study design and development of a preliminary protocol and preliminary patient and caregiver questionnaires.
- Input from cCMVd clinical experts and other stakeholders informed the survey design.
- A conceptual framework communicating the lifetime economic burden of cCMVd informed the general content of the preliminary questionnaires. [Please see the poster "A Framework for Assessing the Lifetime Economic Burden of Congenital Cytomegalovirus in the United States," presented at the 2018 CMV Public Health and Policy Conference September 24, 2018, 4:30 p.m. to 6:00 p.m.]
- A targeted, scientific literature review informed the selection of four validated instruments to collect data on health-related quality of life (HRQOL), productivity, and caregiver burden. The four instruments listed in Table 1 will be evaluated during the qualitative work. In the current phase of work, some gaps were identified and addressed (see next sections).

**Table 1. Validated Instruments Identified for Survey Questionnaire**

Outcome	Survey Measure
<b>Work productivity and activity impairment</b> • Activity impairment due to health problems • Work time missed due to health problems (absenteeism) • Impairment while working due to health problems (presenteeism)	WPAI-GH, de novo questions
<b>Quality of life for adult survey participants (patients and caregivers)</b> • Physical functioning • Role-physical • Bodily pain • General health • Vitality • Social functioning • Role-emotional • Mental health	SF-12
<b>Quality of life for pediatric and adolescent survey participants</b> • Psychosocial • Physical	PedsQL
<b>Caregiver burden</b>	ZBI 12-item scale, de novo questions

PedsQL = Pediatric Quality of Life Inventory; SF-12 = 12-Item Short-Form-Health Survey; WPAI-GH = Work Productivity and Activity Impairment Questionnaire General Health; ZBI-12 = Zarit Burden Inventory 12-item.

## STEPS TO ADDRESS DATA GAPS

- De novo questions for which there is little published data were developed to collect data on participant demographics, out-of-pocket expenditures, caregiver physical and mental health needs, and overall caregiver burden. Table 2 provides some examples of these questions.
- Additional de novo questions may need to be written based on additional qualitative work.

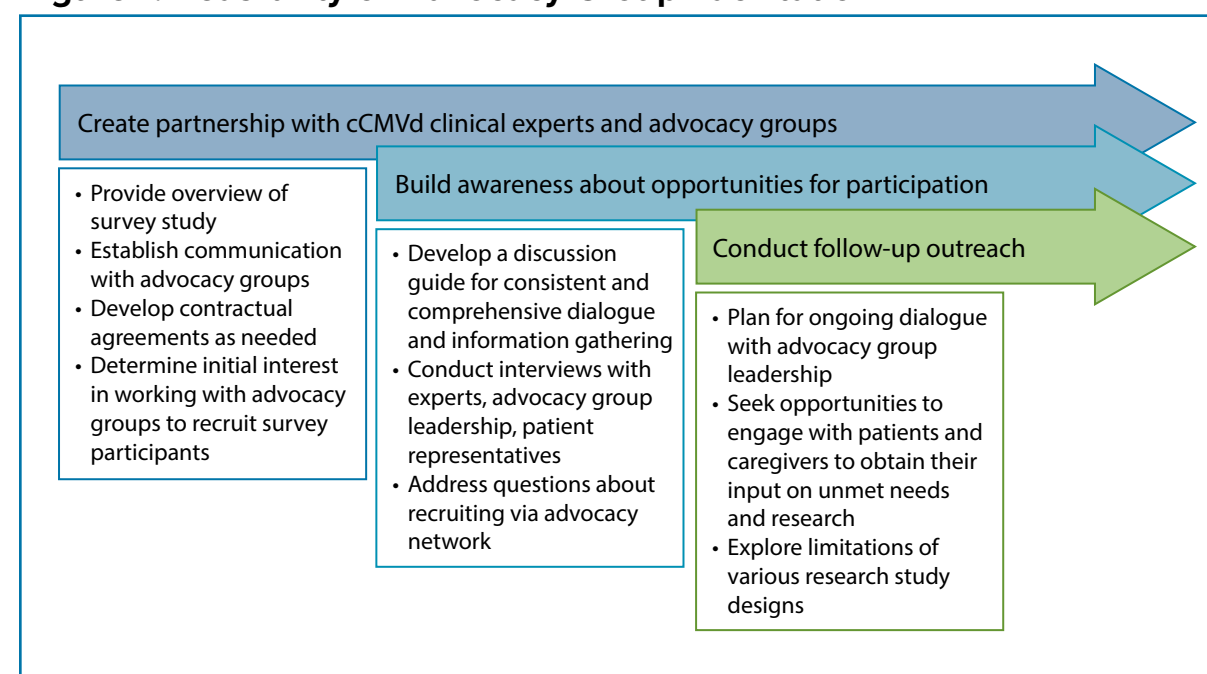
## PROPOSED METHODS FOR ADDITIONAL QUALITATIVE WORK

- Figure 1 illustrates the main activities of a feasibility assessment that will evaluate how advocacy groups could support recruitment of survey participants.
- Additional qualitative research methods illustrated in Figure 2 will be conducted to ensure the questionnaires are developed to collect the necessary data from patients and caregivers to achieve the study objective.

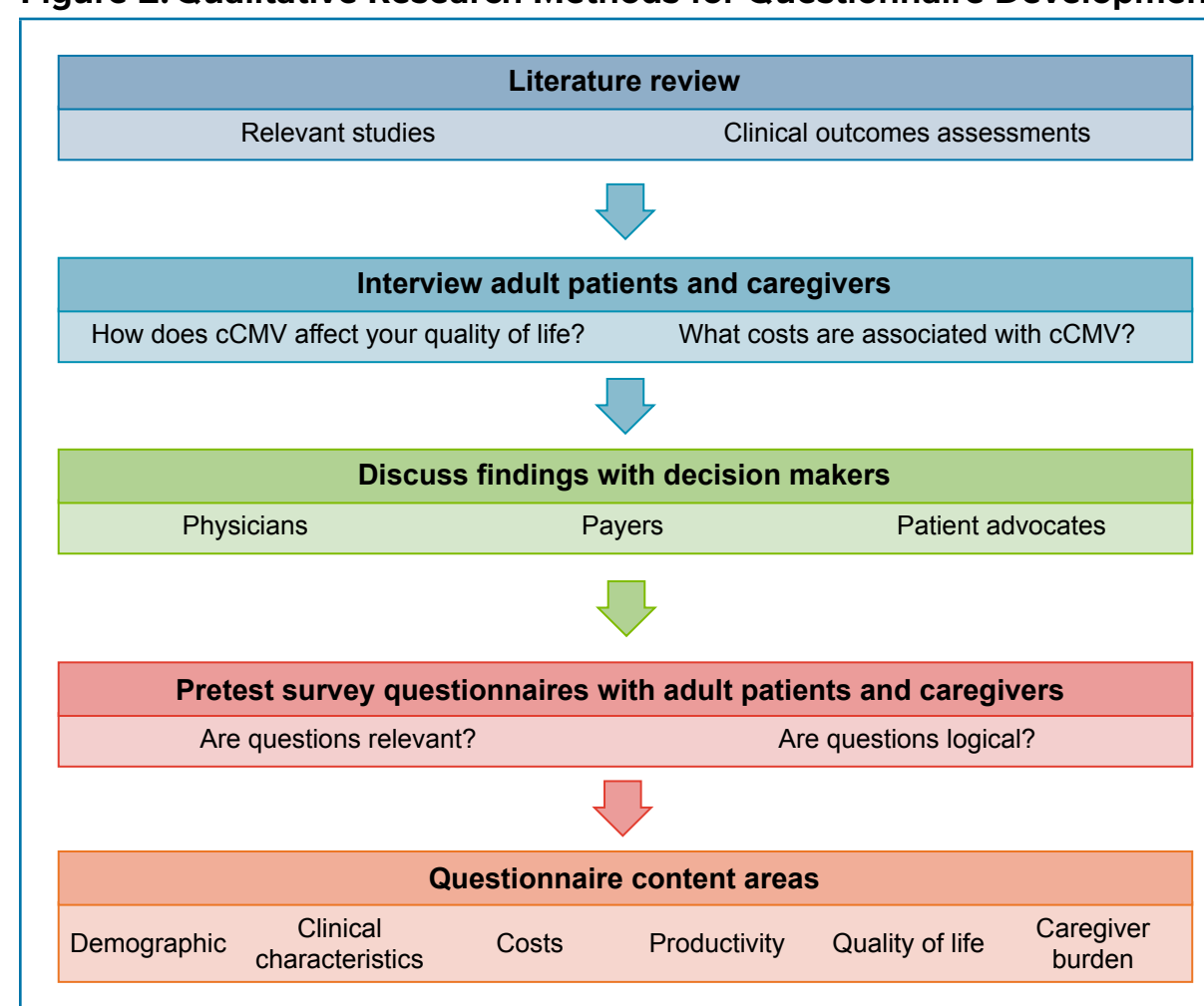
## NEXT STEPS FOR STUDY IMPLEMENTATION

- Upon completion of the feasibility assessment and qualitative work, the design of the survey study will be finalized.

**Figure 1. Feasibility of Advocacy Group Facilitation**



**Figure 2. Qualitative Research Methods for Questionnaire Development**



**Table 2. Examples of Questions Where There Are Gaps in Published Data**

Item	Research Question/Purpose	Survey Questions	Analysis Goal
<b>Objective: To describe indirect and intangible costs among cCMVd caregivers</b>			
Screeners	Enroll adult caregiver for patient with cCMVd	Following referral from advocacy group, confirm cCMVd caregiver	Confirm eligibility
<b>Consent to participate in survey study</b>			
Survey	Demographics	• Age, gender, marital status, education, insurance, household composition	Representativeness of survey population
	Caregiver HRQOL	• During the <u>past 4 weeks</u> , how much of the time has your <u>physical health or emotional problems</u> interfered with your social activities? • During the <u>past 4 weeks</u> , how much <u>did pain interfere</u> with your normal work?	Intangible costs
	Child/adolescent HRQOL	• Questions about how much of a problem the child has with health and activities, getting along with others, school	Intangible costs
	Employment and work productivity	• During the past 7 days, how many hours did you miss from work? • During the past 7 days, how much did your health problems affect your productivity while at work? • Have you or someone in your family ever had to quit working to care for your child with cCMVd?	Indirect costs
	Caregiver burden	• During the past 12 months, how many times have you seen a professional to treat depression, anxiety, or stress? • Do you feel stressed between caring for your child and trying to meet other responsibilities for your family or work?	Indirect costs
	Family out-of-pocket expenses	• Have you had out-of-pocket expenditures for prescription medications, hearing aids, or other medical devices/prosthetics? • Have you had out-of-pocket expenditures that total more than \$300/month for transportation and lodging for cCMVd medical visits?	Direct costs

## CONCLUSIONS

- Evidence on how cCMVd affects the physical, emotional, economic, and social functioning of patients and caregivers is important for policy and health care resource allocation decisions.
- Results from the feasibility assessment and qualitative work will ensure the study design, recruitment plan, and questionnaires are fit for purpose.
- We anticipate this survey will help characterize the economic and humanistic burden experience of those affected by cCMVd.

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