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# A Framework for Assessing the Lifetime Economic Burden of Congenital Cytomegalovirus in the United States

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Congenital Cytomegalovirus  
Public Health & Policy Conference;

September 23-25, 2018;

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## BACKGROUND

- 0.4% to 0.7% of births in the United States (US) are infected with congenital cytomegalovirus infection (cCMV).<sup>1</sup>
- 12.7% with cCMV exhibit disease (cCMVd), and 50% of patients with cCMVd develop permanent physiological abnormalities such as sensorineural hearing loss (SNHL), vision loss, or neurological impairments.<sup>2</sup>
- No US Food and Drug Administration-approved vaccines or medications exist to prevent acquisition of CMV during pregnancy or mother-fetus transmission, but clinical trials are ongoing.<sup>3</sup>
- Decision makers require estimates of cCMV's economic implications to assess the value (e.g., cost-effectiveness) of prevention efforts accurately.

## OBJECTIVE

- Develop a conceptual framework to characterize the lifetime economic burden of cCMV in the US both within and outside the health care (HC) system.
- Identify data gaps to prioritize future research and provide preliminary cost estimates to understand this burden in the US.

## METHODS

- An inventory of cost components (direct HC, direct non-HC, indirect, and intangible costs) associated with cCMVd was developed in accordance with current US cost-effectiveness standards<sup>4</sup> and previous burden-of-illness studies in congenital diseases.<sup>5</sup>

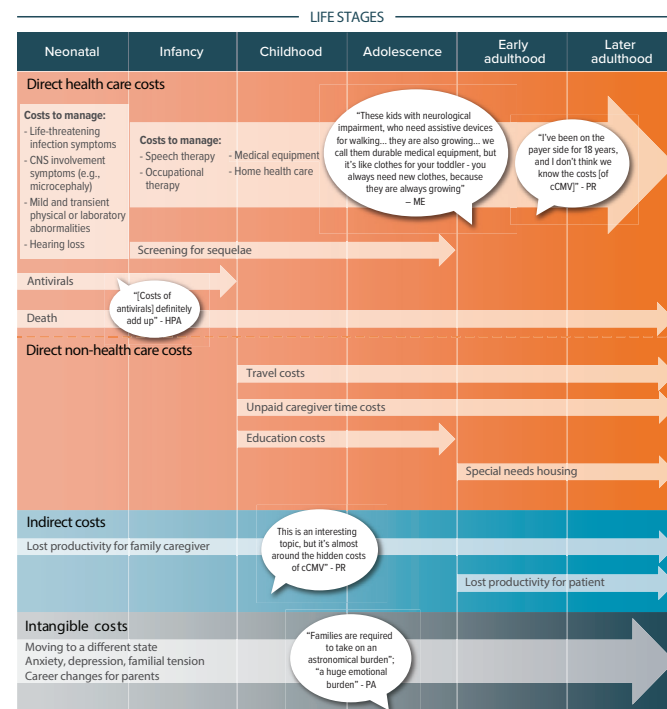
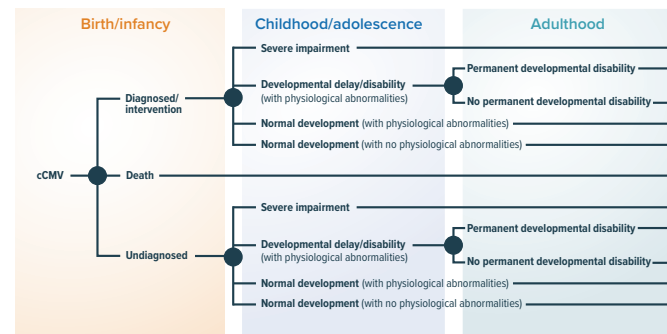
## DISCUSSION

- The lifetime economic burden of cCMV in the US is not well understood in the literature, and substantial data gaps exist for estimating this burden.
- While existing cCMV patient registries may help to fill gaps, challenges exist when using such data (e.g., all cost components and the costs before cCMV diagnosis may not be captured).
- Data from studies of non-cCMV-specific populations (e.g., cost studies of SNHL) may be useful. However, such studies need to include non-HC and indirect costs and measures of the intangible burden of disease.
- This study supports a research agenda in the field of cCMV:
  - Expanding data collected in patient registries (e.g., lost wages, time, and intangible burden of cCMVd)
  - Research into linking or supplementing patient registries with external datasets (e.g., health insurance claims or patient surveys)
  - Estimating the cost of delayed cCMV diagnosis
  - Increasing awareness and population size of patient registries
- Limitations of this study include omitting the economic burden during the prenatal period and using a targeted rather than systematic literature review.
- To accurately evaluate the cost-effectiveness of new interventions for cCMV, research into cCMV's full range of economic consequences on the patient, family, and caregiver needs to be undertaken.

## RESULTS

### Conceptual Framework

Figure 1. Conceptual Framework Chance Tree for Estimating the Lifetime Economic Burden of cCMV



HPA = hospital pharmacy administrator; ME = medical epidemiologist; PA = patient advocate; PR = payer representative.

### Gap Analysis

- 4 cCMV cost studies found<sup>6,7,8,9</sup>
  - All for patients < 1 year of age
  - No non-HC included in any study
- No cost studies focused on patients with severe impairment.
- No studies of the indirect costs or intangible measures of disease burden were found for any patient profile.
- Cost-effectiveness analyses of cCMV interventions mainly rely on data published more than 25 years ago for non-cCMV-specific patient populations with developmental disabilities (e.g., Lawrence et al.,<sup>10</sup> Gantt et al.,<sup>11</sup> and Dempsey et al.<sup>12</sup>

### Data From Targeted Literature Review

- The framework was populated (Tables 1-3) with US-based cost estimates from the literature for patients with cCMV whenever possible.

Table 1. Costs Due to Diagnosis of cCMV in the First Year of Life

Cost Component	Cost Estimate
<b>Direct health care costs per patient</b>	
Post-birth to 1 year old	\$ 50,025 <sup>a</sup>
<b>Direct health care costs per hospitalization</b>	
Birth	\$ 99,978 <sup>b</sup>
	Vaginal: \$ 40,771 <sup>b</sup>
	Caesarian: \$95,853 <sup>b</sup>
Birth to 1 year old	< 1 year old: \$ 76,965 <sup>b</sup>
	< 1 month old: \$ 92,681 <sup>b</sup>
	\$ 106,948 <sup>b</sup>

Table 2. Cost Components for Normal Development with Physiological Abnormality

Cost Component	Annual Cost Estimate
First-year hearing loss is identified	\$1,931 - \$1,991 <sup>11</sup>
< 6 years old	\$1,907 - \$1,956 <sup>11</sup>
6 to 12 years old	\$1,583 - \$1,670 <sup>11</sup>
13 to 17 years old	\$1,574 - \$1,660 <sup>11</sup>
≥ 18 years old	\$977 <sup>11</sup>
Blindness/vision loss	\$6,884 <sup>13</sup>
Retinal disorders (without diabetes)	\$3,854 <sup>13</sup>
Strabismus	\$2,443 <sup>13</sup>

Table 3. Cost Components for Developmental Disability in Childhood/Adolescence and Permanent Developmental Disability in Adulthood

Cost Component	Annual Cost Estimate
<b>Direct HC Costs for Developmental Disability in Childhood/Adolescence</b>	
OOP	ASD: \$217 <sup>14</sup>
	ASD: \$3,602 <sup>14</sup>
Third-party payers	ID without CP: \$25,086 <sup>15</sup>
	CP without ID: \$24,634 <sup>15</sup>
	CP with ID: \$63,847 <sup>15</sup>
<b>Direct Non-HC Costs for Developmental Disability in Childhood/Adolescence</b>	
Education costs	ASD: \$10,269 <sup>14</sup>
Condition-related therapy and family-coordinated services	ASD: \$417 <sup>14</sup>
Unpaid caregiver time costs	ASD: \$6,070 <sup>14</sup>
<b>Direct HC Costs for Permanent Developmental Disability in Adulthood</b>	
OOP	\$2,240 <sup>16</sup>
Third-party payers	\$16,144 <sup>16</sup>

cCMV = congenital cytomegalovirus; OOP = out-of-pocket; ASD = autism spectrum disorder; CP = cerebral palsy; ID = intellectual disability; USD = United States dollars.

Cost estimates are inflated to 2018 USD using the medical care component of the CPI.<sup>17</sup>

## REFERENCES

See handout for references.

## SOURCE OF SUPPORT

This study was funded by Merck Sharp & Dohme Corp., a subsidiary of Merck & Co., Inc., Kenilworth, NJ, USA.

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