

TASK Technology Assessment at SickKids

Dear friends,

Living under the spectre of COVID during the last two years has affected every aspect of our lives profoundly. While for the most part the physical health of children has not been as adversely affected by COVID as others, children may have paid the greatest price in terms of interrupted education, stressful home environments and detrimental emotional effects. Long after the pandemic has ended, the impact on children will persist. It is as critical as ever that we apply methodologic rigour as we attempt to model resource use and health outcomes of children over the long term and understand fully the multi-faceted effects of COVID on the lives of children.

We welcome you to read the papers included in a special collection in *Children* on [Assessing Value in Child Health](#) edited by Dr. Wendy Ungar.

Stay hopeful and healthy -

Wendy



Cost-effectiveness of genome sequencing in pediatric populations

Genome-wide sequencing (GWS) can positively impact the health of children with rare diseases by reducing the time to diagnosis and enabling early intervention. As GWS becomes increasingly available as an option in patient management, understanding its full value and range of benefits to providers, patients and families is critical. Our work at TASK comparing the microcosts of GWS in children with neurodevelopmental conditions to children with cardiac conditions has recently been published in [Genetics in Medicine](#). A team at TASK led by Dr. Robin Hayeems and Dr. Wendy Ungar is studying the [Ontario-wide implementation of clinical GWS](#) through the collection of process and outcomes data and estimation of diagnostic yield for diverse patient groups. The incremental cost-effectiveness of GWS will be studied in patients randomly assigned to receive whole genome or exome sequencing. A sub-study is examining the costs and effects of disclosure of secondary findings. The study is currently enrolling families of children with rare diseases at SickKids and Children's Hospital of Eastern Ontario (CHEO).

Neurodevelopment disorders: services, costs and economic evaluation

TASK completed a cost analysis for a [randomized controlled feasibility trial of a parent coaching intervention for toddlers with suspected autism](#). Results have been [presented](#) to stakeholders and a publication is forthcoming.

Data on service use and costs for autistic adolescents are scarce. TASK is examining service use and cost data as part of the [Phase III of Pathways in ASD](#).

[The CHILD-BRIGHT Network](#) is a pan-Canadian collaboration between children's hospitals across Canada to study novel interventions in children with lifelong brain-based developmental disabilities. TASK, in collaboration with Dr. Jennifer Zwicker at University of Calgary and Dr. Myla Moretti at the SickKids-based Ontario Child Health Support Unit, are conducting cost-effectiveness analyses using patient-level data from multiple [randomized controlled trials](#).

Spillover Effects

Ramesh Lamsal, TASK PhD candidate, is nearing completion of his thesis research on developing theory and methods to incorporate family spillover costs and health consequences in the cost-effectiveness analysis of child health interventions. This work was presented orally at the 2021 annual meetings of the International Health Economics Association and the [Canadian Agency for Drugs and Technologies in Health](#).

Meanwhile the work of former TASK trainee Alex Cernat, MSc on the methodologic

challenges in incorporating cascade effects of genetic testing in economic evaluation is described in two new publications:

- Cernat A, Hayeems RZ, Ungar WJ. Cascade health service use in family members following genetic testing in children: A scoping literature review. *European Journal of Human Genetics* 2021; 29(1): 1601-1610, <https://doi.org/10.1038/s41431-021-00952-4>
- Cernat A, Hayeems RZ, Prosser LA, Ungar WJ. Incorporating cascade effects of genetic testing in economic evaluation: A scoping review of methodological challenges. *Children* 2021; 8(5): 346, <https://doi.org/10.3390/children8050346>

PEDE Update

As part of the [PEDE Project](#), TASK maintains a database of pediatric economic evaluations published since 1980. The database has recently been updated with 2020 publications and now contains detailed information on over 4,000 studies, including an inventory of utility weights. Access to the on-line [database](#) is free and the database is easily searchable. Please contact us if you are interested in collaborating on a study using the PEDE database.

Announcements

In 2021, University of Toronto Institute of Health Policy, Management and Evaluation PhD student Afua Asare successfully defended her doctoral dissertation, *Province-wide vision screening to detect amblyopia and refractive errors in young children enrolled in Ontario's schools*.

In 2021, University of Toronto Institute of Health Policy, Management and Evaluation PhD student Christina Belza successfully defended her doctoral dissertation, *Caregiver burden in children with intestinal failure dependent on long-term parenteral nutrition*.

Awards

The following grants have been awarded:

- Canadian Institutes of Health Research Project Grant. *Genome-wide Sequencing: Secondary Findings Impact Study (SF-Impact Study)*. RZ. Hayeems (PI), WJ. Ungar (co-PI) and co-investigators.
- Genome Canada Genomic Applications Partnership Program (GAPP) in partnership with Ontario Ministry of Health: *Optimization and implementation of a clinical genome-wide sequencing service for rare disease diagnosis in*

Ontario. K. Boycott (PI), B. Reib (PI), R. Hayeems (Evaluation lead), WJ. Ungar (Health Economics lead) and co-investigators.

New Students

TASK welcomes new students:

- Roaa Shoukry, MSc Candidate University of Toronto Institute of Health Policy, Management and Evaluation will conduct a cost-effectiveness analysis of a cognitive rehabilitation program for children with ADHD and ASD.
- Aranie Vijayaratnam, MSc Candidate, University of Toronto Institute of Health Policy, Management and Evaluation will conduct a cost effectiveness analysis of a parent coaching program for children with neurodevelopmental disabilities.

Workshop, Conference and invited presentations

In January 2022, Dr. Wendy Ungar, Dr. Myla Moretti and Kate Tsiplova presented a workshop on Economic Evaluation in Child Health for the Canadian Agency for Drugs and Technologies in Health.

In 2021, Dr. Ungar was the keynote speaker at the Health Technology Assessment Symposium for Economic Evaluation of Exome and Genome Sequencing in Child Health, Center for Drug Evaluation, Taiwan; was an invited speaker on [Economic evaluation of exome and genome sequencing in child health: Tracing the sequence from microcosting to implementation](#) for the Canadian Agency for Drugs and Technologies in Health; and was an invited speaker at the [Symposium on Supporting Early Social & Communication Development through Parent Coaching](#) for the Autism Community Training program. Dr. Ungar was invited to participate in the NICE International – ISPOR Joint HTA Roundtable on Pediatric Health Related Quality of Life and the European Health Economics for Personalized Medicine (HecoPerMed) workshop on Personalised Medicine Specific Health Economic and Payment Modelling.

TASK team members and collaborators presented their projects at numerous 2021 virtual meetings. Highlights include:

- European Virtual Conference on the Diffusion of Genomic Medicine, Dijon, France
 - Ungar WJ, Jegathisawaran J, Tsiplova K, Marshall CR, Stavropoulos DJ, Pereira S, Thiruvahindrapuram B, Hayeems R, Liston E, Reuter M, Manshaei R, Cohn I. Accurate and comprehensive microcosting of genome sequencing in pediatric populations.
 - Ungar WJ, Sikich N, Carroll J, Ng V, McDowell S, Yi J. Health technology assessment and funding of genome medicine technologies in Ontario, Canada.
 - Ungar WJ, Cernat A, Hayeems R. Family Matters: Measuring the preferences of family members for genome sequencing.

- Ungar WJ. Considerations for Cost-Effectiveness Analysis of Genome Sequencing.
- International Society for Autism Research Virtual Annual Meeting
 - Ungar WJ, Tsiplova K, Jegathisawaran J, Karen Kalynchuk. The PACE coaching cost analysis: An incremental analysis comparing the costs of parent coaching to assessment and monitoring in toddlers at risk for ASD.
 - Tsiplova K, Ungar WJ, Szatmari P, Cost K, Pullenayegum E, Duku E, Volden J, Cleve J, den Otter J, Miranda P, Smith IM, Waddell C, Zwaigenbaum L, Bennett T, Elsabbagh M, Georgiades S, Zaidman-Zait A, Kerns C. Measuring the association between behavioural interventions and outcomes in young children with autism spectrum disorder.
- Canadian Agency for Drugs and Technologies in Health Technology Virtual Annual Symposium
 - Ungar WJ, Sikich N, Chakraborty P, Ho C, Hayeems R. Funding and implementation decision-making for genomic medicine technologies
 - Lamsal R, Rahman L, Yeh EA, Pullenayegum E, Ungar W. A theoretical framework for the inclusion of family spillover effects in pediatric economic evaluation.
 - Subasri M, Arje D, Gauvreau C, Wight L, Ungar WJ, Denburg A, Hayeems R. Towards a framework to guide the prioritization, evaluation, and implementation of precision child health technologies: Results from a scoping review.

[TASK WEBSITE](#)

Contact us

Navpreet Kaur
416-813-7654 ext. 309510
navpreet.kaur@sickkids.ca