Economic justification for neurodevelopmental support for children with congenital heart disease

A scoping review of economic-modelling and recommendations for future practice

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Background

Neurodevelopmental delay or disability (NDD) is a significant concern in the care of children across a range of paediatric specialties, including children with congenital or childhood heart conditions.

To date, there has been no attempt to report on the existing decision analytic models with a view to assessing their usefulness for informing healthcare resource allocation decision-making in this context.

The aim of this review was to conduct a scoping review of current economic modelling regarding care for children with, or being monitored for, common neurodevelopmental disorders.

Methods

Stage 1
Identifying the research question

Multidisciplinary panel (n=8) determined the overarching research question:

What model parameters and structures have informed decision-analytic models developed for economic evaluations of care for children with common neurodevelopmental disorders?

Stage 2
Identifying relevant studies

Four electronic databases were searched:

- *PubMed*
- *PsycINFO*
- *International Network of Agencies for Health Technology Assessment*
- *Paediatric Economic Database Evaluation*

Stage 3
Study selection

Inclusion criteria:

- Intervention, or surveillance of children with any of 8 common neurodevelopmental disorders
- Economic evaluation models
- Time horizon 12+ months
- Written in English
- Published since 2000

Exclusion criteria:

- Non-model-based economic evaluations
- Model-based economic evaluation of screening programmes
- A protocol, narrative review, letter, commentary, news article, or conference abstract.

Stage 4
Data collection

Two authors extracted data, including:

- Study characteristics
- Model design (perspective, intervention, comparator, discount rate, model type, time horizon, input parameters, effectiveness measure, sensitivity analysis, and willingness to pay threshold value);
- Health states, utility values and sources
- Information on cost-effectiveness analysis

Stage 5
Data summary and synthesis of results

Summary tables and narrative syntheses were prepared to provide an overview of studies including key study characteristics and methods

Findings were also synthesised for each diagnostic categories.

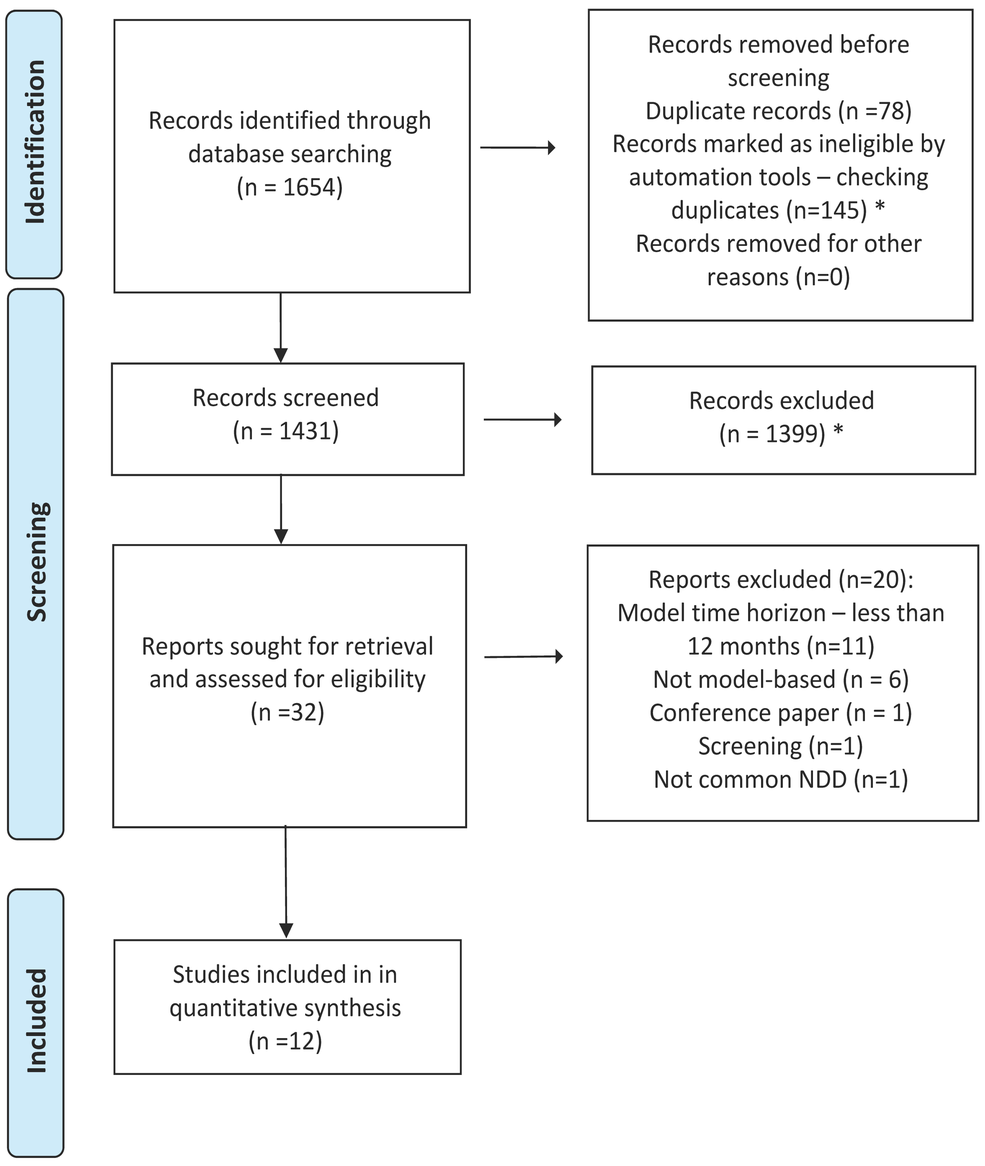
Quality appraisal of reporting

The study reporting quality was assessed using the 24-Item Consolidated Health Economic Evaluation Reporting Standards (CHEERS) statement.

Results

1654 records were identified through database searching –12 studies were included in the final quantitative analysis (Figure 1, Table 1).

Figure 1: PRISMA Flow Diagram – Identification of studies via databases



**Conducted using Rayyan application*

Table 1: General Characteristics of the included studies (n=12)

Item	Specification	Number of studies
Study country	The Netherlands	4
	USA	3
	Canada	2
	Brazil, Spain, UK (each)	1
Type of economic model	Decision-tree	3
	Markov model	6
	Decision tree AND Markov model	3
Study perspective	Societal only	7
	Health care system only	2
	Provincial/Government/Public and Societal	2
	Health Care System, Public Sector, Societal	1
Model time horizon	5-10 years	6*
	11-20 years	5*
	More than 21 years	2
	Lifetime	1
Neurodevelopmental Disorder (NDD)	ADHD	6
	ASD	3
	Cerebral palsy	2
	Dyslexia	1
Intervention classification	Clinical management or follow-up	5
	Pharmaceutical agent	4
	Behavioural program/management	3
Health outcome	Quality Adjusted Life Year (QALY)	9
	Disability Free Life Year (DFLY)	2
	LY of serious delinquent behavior prevented	1

** one study ran model over 6, 12, and 18 years so total exceeds 12;** different discount rates for costs and effects or discounted costs only*
***ADHD**=Attention Deficit Hyperactivity Disorder; **ASD**=Autistic Spectrum Disorder; **QALY**=Quality Adjusted Life Year; **DFLY**= Dependency Free Life Years; **LY**=Life Years*

Key conclusions: There are currently no congenital heart disease-specific economic modelling studies for informing public resource allocation for neurodevelopmental support for children with congenital heart disease. This is a priority for future research in the field.

Economic modelling studies in this field would likely benefit from adopting long time-horizons and considering third-party/family impacts.

While economic analyses in this field are currently scarce, emergent data from common neurodevelopmental disorders was encouraging in the quest for cost-effective care that improves quality of life among these conditions and was often found to be cost-saving.

Source: Kularatna S, Jadambaa A, Senanayake S, Brain D, Hawker N, Kasparian NA, Abell B, Auld B, Eagleson K, Justo R, McPhail SM. 2022. The Cost of Neurodevelopmental Disability: Economic review of current models of care. *(Manuscript under review)*

AusHSI Australian Centre for Health Services Innovation
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CHD LIFE+ Congenital Heart Disease - Long-term Improvement in Functional Health+ Family-centred care models supporting long-term neurodevelopment
An NHMRC MRFF Congenital Heart Disease Grant (ARGCHDG0035)