Screening for malignancy after augmentation cystoplasty in children with spina bifida: a decision analysis
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Record Status
This is a critical abstract of an economic evaluation that meets the criteria for inclusion on NHS EED. Each abstract contains a brief summary of the methods, the results and conclusions followed by a detailed critical assessment on the reliability of the study and the conclusions drawn.

CRD summary
This study assessed the cost-effectiveness of annual screening by cystoscopy and cytology to detect bladder malignancy following augmentation cystoplasty in paediatric patients with spina bifida. The authors concluded that screening by annual cystoscopy and cytology was very unlikely to be cost-effective at accepted willingness-to-pay thresholds. The methods were valid and various areas of uncertainty were considered, supporting the authors’ conclusions.

Type of economic evaluation
Cost-effectiveness analysis, cost-utility analysis

Study objective
This study assessed the cost-effectiveness of annual screening by cystoscopy and cytology to detect bladder malignancy following augmentation cystoplasty (bladder reconstruction) in paediatric patients with spina bifida.

Interventions
Screening by cystoscopy and cytology in addition to usual care was compared with usual care alone, which was an annual physician visit with an ultrasound examination of the kidneys and augmented bladder. Annual screening began 10 years after cystoplasty.

Location/setting
USA/out-patient.

Methods
Analytical approach:
The analysis was based on a Markov model, with a hypothetical cohort of 10-year-old children and a lifetime horizon. The authors stated that a societal perspective was adopted.

Effectiveness data:
A systematic literature review was performed to identify the relevant inputs for the model. No clinical trials were found; the authors selected point estimates from those studies that were found. The incidence of malignancy after cystoscopy was from US cohort studies. Mortality was from the US Centers for Disease Control and Prevention, National Vital Statistics. The efficacy of screening in identifying malignancy was a key input to the model. This was estimated by the authors due to a lack of good published data.

Monetary benefit and utility valuations:
The utility values for the sensitivity analysis were from published sources and authors’ opinions.

Measure of benefit:
Life-years and quality-adjusted life-years (QALYs) were the summary benefit measures. A 3% annual discount rate was applied.

Cost data:
The economic analysis included the costs of annual screening, cystectomy, chemotherapy, cancer follow-up, and end-of-life care. Patient and caregiver time lost was considered. The costs of screening were from the Centers for Medicare
and Medicaid Services. Time was valued using official wages. The costs of the management of cancer were from published studies conducted in the USA. All costs were in US dollars ($) and a 3% annual discount rate was applied. The price year was 2010.

Analysis of uncertainty:
A Monte Carlo micro-simulation used randomly selected combinations of parameter values to assess the overall uncertainty. One- and two-way sensitivity analyses were carried out to determine the parameter thresholds at which the two strategies were equally cost-effective.

Results
The expected costs were $9,780 with usual care and $22,790 with screening. The life-years were 23.47 with usual care and 23.52 with screening. The incremental cost per life-year gained with screening, over usual care, was $273,718, which was well above the cost-effectiveness threshold of $100,000 per life-year gained.

These results were quite stable and screening never approached the cost-effectiveness threshold, except in extreme scenarios, such as if the annual rate of cancer development was more than 2.5 times higher than the base-case estimate or if the test cost was very low. Screening was cost-effective in 11% of the Monte Carlo simulations (3% when QALYs were used).

Authors’ conclusions
The authors concluded that annual cystoscopy and cytology to screen for malignancy in patients with spina bifida after cystoplasty was highly unlikely to be cost-effective at accepted thresholds.

CRD commentary
Interventions:
The selection of the comparators was appropriate as the proposed screening was compared against usual care. The authors pointed out that many questions remained about how suitable the methods were for screening.

Effectiveness/benefits:
The published literature was systematically searched to identify the relevant sources of evidence, but few details of this literature review were presented and the design and key characteristics of the studies found were not reported. Some epidemiological data were from cohort studies and local databases, which should be appropriate for these parameters. No valid data on screening accuracy were available, and the authors estimated a base-case value, which was extensively varied in the sensitivity analysis. This was acknowledged as a limitation of the analysis. The potential heterogeneity among studies was not discussed. Expected survival was an appropriate benefit measure to capture the impact of the disease on the patients’ health. Quality-of-life adjustments were made in the sensitivity analysis, due to the uncertainty around these estimates, and the findings did not favour screening.

Costs:
The categories of costs reflected the societal perspective and included those borne by third-party payers, patients, parents, and caregivers. A clear description of the cost categories was given but they were presented as totals, without their unit costs and resource quantities. The sources for some costs were not fully described, but they generally reflected the US context. An extensive sensitivity analysis was conducted on all the cost parameters. Details, such as the price year and discount rate, were given.

Analysis and results:
The results were extensively presented. The costs and benefits of the strategies were synthesised using an incremental approach. The uncertainty was extensively investigated, using both deterministic and probabilistic analysis, to consider different areas of variability. A Markov model was appropriate for representing the history of disease. The authors acknowledged the need for several assumptions for the clinical and the economic values; they generally used estimates that favoured screening, but still found it was not good value for money. The results should be considered to be specific to the USA and their transferability was not discussed.

Concluding remarks:
The methods were valid and various areas of uncertainty were considered, supporting the authors’ conclusions.

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