Patients with polycystic kidney disease would benefit from routine magnetic resonance angiographic screening for intracerebral aneurysms: 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.

Health technology
Magnetic resonance angiographic imaging (MRI) screening in patients with autosomal dominant polycystic kidney disease (ADPKD).

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
Patients with autosomal dominant polycystic kidney disease (ADPKD) with no history of subarachnoid haemorrhage (SAH) or renal failure.

Setting
The practice setting was hospital. The economic study was carried out in Massachusetts, USA.

Dates to which data relate
Effectiveness data were obtained from studies published between 1949-1992. Resource use data were not reported. Some cost data were obtained from a study published in 1987 (no dates for items of cost data were given). The price year was not stated.

Source of effectiveness data
Estimates for final outcomes were derived from a review of previously completed studies.

Modelling
A Markov model was used to estimate the number of years of life "alive" an ADPKD patient would spend, and also time spent "alive without neurological deficit", and to project costs over lifetime in both the screening and non-screening strategies.

Outcomes assessed in the review
The review assessed autopsy-based and arteriography-based estimates for aneurysm prevalence in ADPKD patients, prospective and population based studies for annual aneurysmal rupture incidence from sub-arachnoid haemorrhage - SAH, and SAH morbidity/mortality rates; risk of cerebral arteriography; MRI sensitivity and specificity for patient with aneurysm; and morbidity/mortality associated with surgical treatment of an aneurysm.
Study designs and other criteria for inclusion in the review
Autopsy-based and arteriography-based studies (for aneurysm prevalence), prospective and population based studies (for annual aneurysmal rupture incidence from sub-arachnoid haemorrhage - SAH, and SAH morbidity/mortality rates), studies investigating risk of cerebral arteriography, MRI sensitivity/specificity and the morbidity/mortality associated with surgical treatment of an aneurysm were searched for.

Sources searched to identify primary studies
Not reported.

Criteria used to ensure the validity of primary studies
Not reported.

Methods used to judge relevance and validity, and for extracting data
Not reported.

Number of primary studies included
A total of 29 studies (1 review, 1 meta-analysis, 1 long term follow-up, 2 prospective studies, others unknown) were directly used as the references for the estimates used in the model, while a total of 35 studies were referred to generally.

Methods of combining primary studies
Not reported.

Investigation of differences between primary studies
Not performed.

Results of the review
(1) Aneurysm prevalence: (a) autopsy-based studies = 14% (95% CI: 11 - 19%); (b) arteriography studies = 16% (95% CI: 10 - 24%); midpoint (15%) selected for analysis.

(2) Annual incidence of aneurysmal rupture = 1.6%.

(3) Morbidity and mortality rates associated with subarachnoid haemorrhage = 70% and 50% respectively.

(4) Risk of transfemoral arteriography = 0.2% (95% CI: 0 - 0.4%).

(5) Morbidity and mortality rates associated with surgical treatment of an unruptured aneurysm = 4.1% and 1.0% respectively.

(6) MRI sensitivity and specificity for patient with aneurysm was 0.95 and 1.0, respectively.

Measure of benefits used in the economic analysis
The benefit measures were ADPKD patient additional life expectancy, and additional years of life without neurological disability.
**Direct costs**
No discounting was performed. Costs and quantities were not analysed separately. The cost items were reported separately. The cost boundary adopted was that of society. Direct costs included: MRA study costs, the cost of cerebral angiography, the costs of surgery and hospitalisation, and caring costs of disability and mortality (from the literature, 1987). Sources of cost data were not given. The date of the price data was not specified.

**Indirect Costs**
No discounting was performed. Costs and quantities were not analysed separately. Lost income from disability and mortality was regarded in the analysis of indirect costs. The cost boundary adopted was that of society. Sources of indirect cost data were not given. The date of the price data was not specified.

**Currency**
US dollars ($).

**Sensitivity analysis**
One and two-way sensitivity analysis varied aneurysm prevalence, annual probability of rupture, SAH morbidity and mortality, operative morbidity and mortality, sensitivity of MRA for aneurysm patients, specificity of MRA for aneurysms, and angiography complications.

**Estimated benefits used in the economic analysis**
Screening would provide 0.9 additional years of life and 1.0 additional year of life without neurological disability to a 20 year old ADPKD patient.

**Cost results**
With an aneurysm prevalence of 15%, the approximate lifetime cost of a screening strategy was $14,000, compared to a non-screening cost of $24,000.

**Synthesis of costs and benefits**
A synthesis of costs and benefits was not required since the MRI screening strategy was the dominant strategy. The cost-effectiveness ratio (incremental cost to additional life years saved) was plotted against aneurysm prevalence. The incremental cost per year of life saved was negative (saving) for a wide range of reasonable values of aneurysm prevalence. The most sensitive parameters of the model were the prevalence of aneurysm in ADPKD, the annual incidence of rupture, and the morbidity and mortality rates associated with rupture.

**Authors' conclusions**
The model predicts that an MRI screening strategy would increase the life expectancy of young patients with ADPKD and reduce the financial impact on society of ADPKD.

**CRD COMMENTARY - Selection of comparators**
The reason for the choice of the comparator is clear.

**Validity of estimate of measure of benefit**
The internal validity of the estimates of the benefit can not be assessed due to the lack of detail regarding the literature review and quality assessment of the primary studies included therein.

**Validity of estimate of costs**
Resource utilisation was not reported separately from the costs. However adequate details of methods of cost
estimation were given. The costs were not discounted despite the lifetime time horizon considered in the cost analysis. Sources of cost data were not systematically reported. No price year was specified, thus hampering cross-study comparisons.

Other issues
In view of the lack of a quality assessment of the primary studies included in the review, the results may need to be treated with some caution.

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