A cost-utility analysis of neonatal circumcision
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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
Routine neonatal male circumcision was assessed.

Type of intervention
Other: Cultural-religious practice; potentially primary prevention.

Economic study type
Cost-utility analysis.

Study population
The study population comprised a hypothetical cohort of healthy male neonates, undergoing routine circumcision or retaining intact genitalia.

Setting
The setting of the study was secondary care. The economic study was conducted in the USA.

Dates to which data relate
The effectiveness evidence was derived from studies published between 1949 and 2000. The cost data, including estimates on resource use, were derived from studies published between 1952 and 1999. The price year was 1999.

Source of effectiveness data
The effectiveness data were derived from a review or synthesis of completed studies.

Modelling
A Markov analytic model was developed to assess the total costs and benefits resulting from routine neonatal male circumcision. The model incorporated health states associated with the intervention and those potentially affected by the intervention. Health states associated with the intervention included immediate complications and the need for a future circumcision revision. Health states potentially affected by the intervention were urinary tract infections, phimosis, balanitis, sexually transmitted diseases (STDs), human immunodeficiency virus (HIV), penile cancer, and so on. The transition probabilities for the health states potentially affected by circumcision were determined for both circumcised males and males with intact genitalia. The timeframe of the analysis was 72 years. The cycle length was one year.

Outcomes assessed in the review
The outcomes assessed included:
cumulative incidence rates of conditions following neonatal circumcision, such as immediate complications, death from the procedure, subpreputial debris, coronal adhesions, meatitis, meatal stenosis, foreskin restoration and circumcision revision;

cumulative incidence rates of other penile-related conditions, such as urinary tract infections in the first year of life, phimosis, balanitis, pyoderma, STDs, HIV and penile cancer, in both circumcised males and males with intact genitalia; and

age distributions of the onset of phimosis, balanitis, STDs, HIV and penile cancer.

Study designs and other criteria for inclusion in the review
Any articles published since 1900 that addressed the impact of neonatal circumcision on health were considered, although preference was given to the most current information. Owing to the lack of randomised controlled trials, observational studies were considered valid for the purpose of the study. No other criteria for inclusion in the review were stated.

Sources searched to identify primary studies
MEDLINE was searched for relevant studies using "circumcision" as a search word. The citations in pertinent articles were also reviewed.

Criteria used to ensure the validity of primary studies
The criteria used were not explicitly reported. Different criteria were used for extracting data on each of the effectiveness parameters examined, based on the availability of relevant studies. However, owing to a lack of trial-based data in most cases, the criteria used were rather crude.

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

Number of primary studies included
Approximately 49 primary studies were included in the review.

Methods of combining primary studies
For most of the parameters examined, the primary studies were combined using narrative methods. In the case of estimation of susceptibility to HIV and STDs of circumcised and non-circumcised males, a meta-analysis of published data was performed using a random-effects model.

Investigation of differences between primary studies
The author considered differences between the primary studies. In some cases, differences in the results between primary studies were attributed to different definitions of a pathological situation, study designs, or age of study populations.

Results of the review
The cumulative incidence rates of conditions following neonatal circumcision were:

immediate complications 3.10%;

death from the procedure 0.0002%;
subpreputial debris 10%;
coronal adhesions 12%, of which 2% required surgical correction;
meatitis 20%;
meatal stenosis 5%, half of which required surgery;
foreskin restoration 0.1%; and
circumcision revision 2.8%, with complications occurring in 5% of cases.
The cumulative incidence rates of other penile-related conditions in circumcised males were:
urinary tract infections in the first year of life 1.01%;
phimosis 1.00%;
balanitis 10%;
pyoderma 6.4%;
bacterial STDs 14.09%;
herpes 1.10%;
warts 3.51%;
HIV 0.0287%; and
penile cancer 0.0290%, with 20% mortality.
The cumulative incidence rates of penile-related conditions in non-circumcised males were:
urinary tract infections in the first year of life 1.52%;
phimosis 0.90%;
paraphimosis 0.05%;
balanitis 8%;
smegma 2%;
pyoderma 2.70%;
bacterial STDs 13.31%;
herpes 1.30%;
warts 3.09%;
HIV 0.0509%; and
penile cancer 0.0870%, with 20% mortality.

Age distributions of the onset of penile-related conditions were not reported in the majority of the cases.
Methods used to derive estimates of effectiveness
In some cases, experts' opinions were obtained so that relevant values could be determined for parameters for which the evidence from the literature review was contradictory or inconclusive.

Measure of benefits used in the economic analysis
The measure of benefits used, expressed as benefit-losses, was the number of well-years lost per 1,000 males resulting from neonatal circumcision compared with non-circumcision. The utilities were estimated using the Quality of Well-being Scale and published values. The benefits were discounted. Two analyses using annual discount rates of 3% (base-case) and 5% were performed.

Direct costs
The direct costs consisted of medical costs only. These included the costs of neonatal circumcision (physician's fee, hospital costs, costs of immediate complications) and costs associated with future penile-related conditions and diseases potentially affected by the procedure in terms of incidence rate, such as urinary tract infections (including further complications such as urosepsis, vesicoureteral reflux and renal disease), phimosis and paraphimosis, coronal adhesions, balanitis, pyoderma, subpreputial debris, circumcision revision, foreskin restoration, STDs, HIV and penile cancer. The costs and the quantities were not reported separately. The costs were derived from literature published in 1984 to 1999. The total costs were derived using modelling. The costs were adjusted to 1999 prices using the Consumer Price Index. They were discounted at rates of 3% (base-case) and 5%, as they were incurred practically over lifetime (72 years).

Statistical analysis of costs
The costs were treated in a deterministically. No statistical analysis of the costs was undertaken.

Indirect Costs
The indirect costs generally referred to the parents' time lost from work for treatment and physician visits. The exception was time off work during hospitalisation of normal newborns, including inpatient stay due to immediate complications following circumcision. In this case no indirect costs were calculated, since one or both parents were thought to take time off work related to the birth of a child. Time lost from work was assumed to equal the length of hospitalisation and/or the length of illness of severity that would prevent return to day care. In the case of a physician visit, time lost from work was assumed to equal 4 hours or half a day of work. Time losses were valued using the May 1999 average earning in the private sector of the USA. The indirect costs were discounted at rates of 3% (base-case) and 5%. Year 1999 prices were used.

Currency
US dollars ($).

Sensitivity analysis
A sensitivity analysis was undertaken to examine the impact of uncertainty arising from variability in the data on the results of the analysis. One-way sensitivity analyses were carried out on the 47 most influential parameters. In addition, analyses using best- and worse-case scenarios were performed. Finally, a Monte Carlo simulation taking 1,000 samples was conducted. This employed the distributions of 24 of the most influential variables. The ranges for the sensitivity analyses were based on either 95% confidence intervals available in the published literature or derived from the meta-analyses undertaken specifically for the study, or on the range of values reported in the studies included in the review.

Estimated benefits used in the economic analysis
The base-case incremental benefit-losses per 1,000 individuals resulting from neonatal circumcision in comparison with non-circumcision were 15,925 well-years lost (undiscounted), 15,300 well-years lost (discounted at 3%), and 14,609 well-years lost (discounted at 5%).
Benefit-losses were calculated over an individual's lifetime. Side effects of treatment, such as immediate and future complications, were considered in the estimation of benefit-losses.

Cost results
The base-case incremental costs per individual resulting from neonatal circumcision in comparison with non-circumcision were $778.66 (undiscounted), $828.42 (discounted at 3%), and $837.59 (discounted at 5%).

The costs were calculated over an individual's lifetime. The costs of adverse events (immediate and future complications from the procedure) were included.

Synthesis of costs and benefits
Routine neonatal male circumcision was more costly than non-circumcision and had an overall negative impact on health. Therefore, non-circumcision was the dominant strategy, and no further synthesis of the costs and benefits was needed. All 47 variables used in the one-way sensitivity analysis had no impact on the results.

Variables related to the initial circumcision were most influential. For all variables, the incidence required to make neonatal circumcision cost-neutral was far outside the realm of what had been reported in the medical literature.

If neonatal circumcision was cost-free, immediate complication-free, had no additional days of hospitalisation, and had no immediate negative impact on health, it would still be more costly (incremental cost $400.03 per individual) and have an overall negative impact on health (incremental benefit-loss 3.34 well-years per 1,000 individuals).

According to the best-case scenario favouring circumcision, this was still more costly than leaving intact genitalia. It provided more benefits only when these were left undiscounted.

The Monte Carlo simulation resulted in an incremental cost of neonatal circumcision of $908.65 per patient (standard deviation $167.80), and an incremental utility in well-years lost per 1,000 individuals of 16.10 (standard deviation 3.81), at a 3% discount rate.

Authors' conclusions
Neonatal circumcision was more costly and had more adverse health effects over the lifetime than forgoing the procedure would result in, regardless of "reasonable" values placed on the variables in the model.

CRD COMMENTARY - Selection of comparators
The comparator of the analysis was the do-nothing option (leaving intact genitalia). This, apparently, represents the only reasonable comparator to routine circumcision.

Validity of estimate of measure of effectiveness
The review of the literature was likely to have been systematic. However, the methodology was only described in outline, so the possibility of introducing biases cannot be excluded. Effectiveness estimates were generally combined using narrative methods. In the case of the estimation of susceptibility to HIV and STDs of circumcised and non-circumcised males, a meta-analysis of published data was performed using a random-effects model. The author investigated and gave possible explanations for the differences between the primary studies when estimating effectiveness. Uncertainty was investigated in a sensitivity analysis.

Validity of estimate of measure of benefit
The estimation of measure of benefits was modelled. The Markov analytic model used for this purpose was appropriate, as it incorporated a large number of relevant health states potentially affected by the intervention. In addition, it allowed the estimation of benefits related to the intervention and its comparator practically over lifetime.
Validity of estimate of costs
It was reported that the analysis adopted a societal perspective, but some categories of costs (e.g. direct personal costs such as travel expenses) were not included in the analysis. Nevertheless, the omission of these costs is unlikely to have affected the results substantially. For the other categories of cost, all relevant cost components were included in the analysis. The costs and the quantities were not reported separately, which this may limit the reproducibility of the results. Sensitivity analyses considered the impact of uncertainty in the cost estimates on the results. Discounting was appropriately undertaken since the costs were incurred over lifetime. The year to which the costs referred was reported, and this increases the generalisability of the results.

Other issues
The author made appropriate comparisons of the findings with those from other studies and found them to be consistent. The issue of generalisability to other settings (Africa) was addressed. The author reported as a limitation of the analysis the poor quality of the vast majority of the studies included in the review, owing to a lack of randomised controlled trials in the area. It was therefore acknowledged that the base-case results were only a rough estimate of the cost-effectiveness of neonatal circumcision, and that the breadth of the confidence intervals reported in the Monte Carlo stimulation reflected more closely the uncertainty in reality. The exclusion of several factors from the analysis, such as neonatal severity of pain, owing to the difficulty in quantifying their effect, was described as another limitation of the study. The author reported the results in full and the conclusions reflect the scope of the analysis.

Implications of the study
The author stated that neonatal circumcision cannot be justified financially or medically, and any justification should be on religious, cultural, or aesthetic grounds. It was recommended that the medical community refrain from promoting, encouraging or presenting neonatal circumcision as a medical option. It was also recommended that third-party payers reassess their reimbursement policies and possibly consider paying physicians or parents not to perform neonatal circumcisions, as both options would result in overall cost-savings.

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