Neonatal Circumcision: Cost-Effective Preventive Measure or "the Unkindest Cut of All"?

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Circumcision, or surgical removal of the penile foreskin, is the subject of a cost-utility analysis appearing in this issue of *Medical Decision Making*.[1] In an Egyptian bas-relief dating back to roughly 4000 BC,[2] one patient appears willing to undergo the procedure. His more reluctant companion seems to require physical restraint, and this divergence of opinion presages the controversies still surrounding this procedure. It is not known to what degree *routine* circumcision (performed as a preventive rather than a therapeutic measure) favorably or unfavorably affects the risks of mechanical, inflammatory, infectious, and neoplastic processes, not to mention sexual sensation in males and their partners. The most serious potential complications of circumcision can be tragic[3] but are fortunately rare. The advisability of this procedure has been addressed in countless letters, case reports, and, of necessity, observational studies of variable methodological rigor. These discussions are far too numerous to even begin recounting here. However, the volume of ink and effort devoted to them affirms that "the history of these few millimeters of skin is utterly epochal and fascinating."[4] Most reviewers concede that there is no conclusive medical evidence of either net benefit or net harm.[5–7]

Neither the American Academy of Pediatrics (AAP)[8] nor the Canadian Paediatric Society (CPS)[9] still recommends circumcision as a routine procedure, with CPS being a bit less enthusiastic. Both bodies recommend that parents be provided with balanced information on the procedure's potential benefits and risks, and both acknowledge that there are religious, ethnic, and sociocultural considerations that tend to drive the final decision anyway.[10–12] If circumcision is performed, it has been recommended that the procedure be accompanied by documented informed consent, as well as by adequate analgesia.

Multistate administrative data from the Federal Healthcare Cost and Utilization Project (HCUP) indicate that 1.2 million males (59% of all US male newborns and 86% of those without a complicating diagnosis) were circumcised at birth in 2000.[13] Figures are felt to be somewhat lower in Canada, and considerably lower elsewhere in the world.[8] It is difficult to isolate costs specifically attributable to circumcisions performed during the birth admission. Nonetheless, the volumes of procedures performed make the aggregate "up front" and potential "downstream" costs (so to speak) of various circumcision strategies an important area to study.

This discussion of Van Howe's article may be prefaced with a review of the few formal cost analyses of circumcision that preceded it. A 1984 Canadian study[14] estimated the mean costs of neonatal circumcision at Can$38.32 per case. Through compounding at 4% annually, this amount would have been worth $272 at age 50. Neonatal circumcision was assumed to prevent the 2 penile cancer cases estimated to occur otherwise per 100,000 50-year-old men per year. The modeled cost was Can$13.6 million per cancer case averted, and the authors concluded that "until demonstrated otherwise, prophylactic neonatal circumcision should be regarded as cosmetic surgery, paid for directly by parents wishing it."

A 1991 cost-utility analysis[15] arbitrarily assigned utilities to death (0), penile cancer (0.5), other penile problems such as phimosis (0.99), and survival without such problems (1.0). No disutilities were assigned

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to circumcision pain or to certain other outcomes. Utilities were combined with literature estimates of the probabilities of neonatal urinary tract infection (UTI), penile cancer, phimosis, and other outcomes. The bases for "cost" estimates ranging from $100 for circumcision to $5000/year for treating penile cancer were not provided. Routine circumcision generated 0.028 more quality-adjusted life years (QALYs) over 85 years, at a mean incremental mean cost of $25.36. Although the "base case" incremental cost-effectiveness ratio of $919.87 per QALY was considered acceptable, it was noted to reflect small and potentially imprecise cost and QALY differences. The authors considered their results to allow parental preference to drive the decision.

A more extensive cost-utility analysis was published almost simultaneously in *Medical Decision Making*.[16] Based on societal quality of well-being (QWB) scores for adults, Ganiats and others assigned disutilities to features such as the inherent pain of neonatal circumcision, symptoms driving future circumcision, procedure complications, UTIs, and penile cancer. These disutility weights, multiplied by symptom durations and by adverse event probabilities based partly on literature estimates, were used to calculate "well-years lost." Billed hospital charges (not costs) were discounted at 5% per year, although costs of physician care may not have been included. Neonatal circumcision was dominated in the base case analysis, with mean incremental costs of $102.45 and incremental losses of 0.0016 well-years per patient. Results were sensitive to some cost and outcome parameter values. The health effect and costs differences were considered sufficiently small to allow the decision to reflect cultural or religious preference.

A more recent study[17] favoring circumcision calculated the 1-year cumulative incidence and mean inpatient/outpatient facility costs (not charges) of treating UTI. Figures for uncircumcised male infants were estimated at 2.15% and $1,179 respectively, versus 0.22% and $703 for circumcised males. As this was acknowledged not to be a comprehensive cost-effectiveness analysis, it was not surprising that others[18–22] using the same data reached differieng conclusions regarding the value of the procedure.

The studies cited above varied in their adherence to principles promulgated since 1977[23–27] to facilitate the generation of methodologically valid, reproducible, transparent, and comprehensive economic analyses. The current cost-utility analysis may be reviewed in this context. In this study, societal perspective costs and quality-adjusted survival associated with circumcising versus not circumcising boys at birth were projected over a 72-year life expectancy. In a Markov analysis with Monte Carlo simulations, QWB scores from Ganiats and others[16] were augmented by values assigned to other outcomes and applied to more recent event probability data. As a key update to the previous studies, estimates of associations between circumcision and the risks of contracting human immunodeficiency virus (HIV) and other sexually transmitted diseases (STDs) were now included. Neonatal circumcision was dominated in the base case analysis, with stated mean incremental (discounted) costs of $828.42 per patient and losses of 15.3 well-years per 1000 patients. Sensitivity analyses were stated to identify no plausible scenarios making neonatal circumcision cost-effective. The author concluded that neonatal circumcision as a medical procedure is not justifiable on financial or medical grounds and that it is not good health policy.

The results of this study indicate that circumcision is not cost-effective from a societal perspective. However, the author's previous research and discussion pieces (some of which are used in this study) do indicate a particular prior perspective on this issue. References 14–16 and 28 cited as having "failed to demonstrate the cost-effectiveness of the procedure" include one study[28] that did not formally address cost issues at all. As described above, baseline results of another study[15] favored circumcision, although minimally so. Authors of this study and of an analysis[16] in which circumcision was dominated both considered their results to be equivocal. As the other study cited[14] only addressed penile cancer issues, its conclusions cannot be considered those of a comprehensive cost-effectiveness analysis. An analogous study[17] that favored circumcision as a means to avoid urinary tract infections and their attendant costs was not mentioned in the current study.

The circumcision "cost" figures used in this study may overestimate the marginal increase in costs or combined parent/insurer reimbursement associated with adding circumcision to the inpatient care already being provided to newborns. Presumably there are more recent figures than the inflation-adjusted physician reimbursement survey data from 1993 to 1994 used in this study. Adding 82% to the physician reimbursement in order to estimate US hospital costs may be questionable. This calculation reflects 1979 to 1983 data cited above[14] that combined Ontario provincial physician *reimbursement* of Can$21.25 with estimated average (rather than incremental) hospital *costs* of $17.07 for nursing personnel time, equipment, supplies, and facility use. Although a source for UTI management costs is noted,[29] no basis is provided for other

care cost figures. It is unclear whether or not the listed estimates include costs of physician care. Any reliance on charges rather than costs could overestimate apparent hospital costs, although it is unclear whether or not the listed estimates include costs of physician care.

Assessments of circumcision effects on rates of UTIs, STDs, HIV, penile cancer, and other conditions have been acknowledged to be complicated by methodological issues such as differential distribution of factors such as prematurity, breastfeeding, "rooming-in" status, use of variable diagnostic tests, recall bias, socioeconomic factors, variable duration of follow-up, variations in sexual practices, misclassification of circumcision status, and uncertain generalizability of international results to US populations. Van Howe notes that few observational studies used in this analysis considered, much less adequately controlled for, such factors. As a result, many frequency rates used in the study are open to question. Consequently, as an example, calculations such as those indicating that circumcised men are at a slightly increased risk of contracting STDs require stronger documentation than a 1999 review article published by the author.[30]

The extensive data that are provided in this study could be presented with more consistency. As mean incremental costs are expressed per individual, the well years lost (0.0153) should also be expressed per individual (rather than per 1,000 individuals). Some "cumulative incidence" data in Table 1 do represent lifetime risks per patient, whereas other frequency figures (e.g., % with urosepsis) are proportions of patients within a given subgroup (e.g., UTI patients). Furthermore, to allow assessment of the relevance of the data to current US populations, Table 1 could have included literature references and/or characterizations of data sources by years (ideally of data collection rather than publication), as being from developed v. developing countries, and so on. This would alert the reader to the fact that available data used to estimate circumcision mortality rates were published in 1949 [31] and 1974,[32] for example.

The bases for the previously published disutility weights[16] used in this study are themselves open to question. For example, it is unclear whether or not the figure of 0.349 for circumcision pain assumes that analgesia was used. Given the subjective nature of disutility weights, it would also have been important for the author to have provided some basis for the weights he generated himself. In addition, this study did not formally consider the fact that some males might assign some disutility to circumcision status (in either direction) that is culturally or religiously dissonant relative to that of family members or peers. Sensitivity analyses could have been used to model the impact of a wide potential range of such disutilities. Given the minimal difference in medically oriented utilities calculated for the 2 strategies, consideration of nonmedical disutilities (especially those experienced over significant portions of one's lifetime) could have significantly affected the analytic results. The inclusion of such considerations is not intended to trivialize the discomfort of "short term" outcomes such as painful circumcision. However, the impact of transitory events on calculated "well years lost" may be limited by their short duration.

There are no available data on the disutility that others might ascribe to the secondary effects of circumcision (e.g., on parental satisfaction or on the transmission of HIV or STDs to sexual partners of circumcised or uncircumcised males). Including such effects also would complicate attempts to compare these results to those of other studies. For these reasons, it was reasonable to formally exclude such considerations from this study.

Thus, through an extensive literature review and generally acceptable methods, this study updates projections of potential health outcomes relative to dollars spent for routine circumcision. It demonstrates the absence of compelling justification for this procedure. However, it is not clear to what degree this may be considered as affirmative evidence of a *lack* of benefit. This uncertainty derives in part from the study's limited methodological transparency, its reliance on understandably suboptimal data, and the fact that it does not address some issues important to the circumcision debate.

So where do we go from here? We should remember that cost-utility analysis may help *guide* health policy decisions, but it does not *make* them. The final decision should consider how the impact of costs and benefits projected for a given policy choice compare with those for alternative uses of the same resource. As may be especially true in the case of circumcision, such deliberations should explicitly consider the importance of various sociocultural and other issues that may not be fully captured in quantitative analyses. Those who consider the current analysis to be incomplete may wish to perform further work to address various methodological issues and to more fully capture the potential impact of circumcision on individuals and/or society as a whole. For those who consider Van Howe's results to be compelling, the next step may be to address explicitly the degree to which finite societal health care resources should be expended for a procedure performed primarily for sociocultural and religious reasons.

Among our neighbors to the north, Manitoba is the only Canadian province that still paid for routine circumcision as of 2002.[33] At least in the short term, elimination of the payment benefit in Ontario in the mid-1990s seemed to have had little effect on circumcision rates in referral centers there.[34] However, as 2002 reimbursement in Manitoba was only Can$19.50,[33] Ontario parents would probably have been relatively insensitive to similar financial disincentives associated with "de-listing" circumcision in that province.

In 2002, Arizona became the 7th state to deny Medicaid funding for nontherapeutic neonatal circumcisions.[35] Van Howe acknowledges that marketing considerations and other factors might still compel US private insurers to continue paying for the procedure. Such dichotomy in reimbursement policies might raise ethical and other concerns that would have to be addressed, including the potential for "two-tiered" access to services. In this context, it might also be of interest to model the outcome/utility changes, to be combined with amounts that parents would need to be willing to pay out of pocket, that might make neonatal circumcision (and its entrained effects) cost-neutral or at least incrementally cost-effective by current standards to the parties that fund the US health care system.

It is the author's prerogative to conclude with a suggestion that insurance companies consider *paying* physicians and parents *not* to participate in or allow performance of neonatal circumcision. However, this is a rather extreme approach that seems to impugn the motives of both groups. Van Howe's assertion that "Either of these options would result in an overall cost savings (sic)" must also be regarded as speculative at this point. A 2003 Cochrane review[36] indicates that "researchers' personal biases and the dominant circumcision practices of their respective countries" complicate interpretation of the existing data on the effect of circumcision on HIV transmission rates. One hopes that future discussions will reflect dispassionate deliberation, rather than the polarized proclamations that have often characterized the entire circumcision debate thus far.

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