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A critical assessment of recent evidence

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Published in: Global Public Health

DOI (link to publication from Publisher): 10.1080/17441692.2016.1184292

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Publication date: 2018

Document Version Publisher's PDF, also known as Version of record

Link to publication from Aalborg University

Citation for published version (APA):

Frisch, M., & Earp, B. D. (2018). Circumcision of male infants and children as a public health measure in developed countries: A critical assessment of recent evidence. Global Public Health, 13(5), 626-641. https://doi.org/10.1080/17441692.2016.1184292

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Global Public Health



An International Journal for Research, Policy and Practice

ISSN: 1744-1692 (Print) 1744-1706 (Online) Journal homepage: http://www.tandfonline.com/loi/rgph20

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To cite this article: Morten Frisch & Brian D. Earp (2016): Circumcision of male infants and children as a public health measure in developed countries: A critical assessment of recent evidence, Global Public Health, DOI: 10.1080/17441692.2016.1184292

To link to this article: https://doi.org/10.1080/17441692.2016.1184292

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Circumcision of male infants and children as a public health measure in developed countries: A critical assessment of recent evidence

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ABSTRACT

In December of 2014, an anonymous working group under the United States' Centers for Disease Control and Prevention (CDC) issued a draft of the first-ever federal recommendations regarding male circumcision. In accordance with the American Academy of Pediatrics' circumcision policy from 2012 - but in contrast to the more recent 2015 policy from the Canadian Paediatric Society as well as prior policies (still in force) from medical associations in Europe and Australasia – the CDC suggested that the benefits of the surgery outweigh the risks. In this article, we provide a brief scientific and conceptual analysis of the CDC's assessment of benefit versus risk, and argue that it deserves a closer look. Although we set aside the burgeoning bioethical debate surrounding the moral permissibility of performing non-therapeutic circumcisions on healthy minors, we argue that, from a scientific and medical perspective, current evidence suggests that such circumcision is not an appropriate public health measure for developed countries such as the United States.

ARTICLE HISTORY

Received 23 October 2015 Accepted 23 April 2016

KEYWORDS

Male circumcision; international public health; health policy; HIV prevention

Introduction

Male circumcision is the surgical removal of part or all of the penile foreskin (Cold & Taylor, 1999). When circumcision is performed on children in the absence of penile disease, it raises a number of complex ethical issues, some of which have been discussed in the recent literature (e.g. Darby, 2015; Earp, 2015a, 2015b; Foddy, 2013; Frisch et al., 2013; Mazor, 2013; Munzer, 2015; Savulescu, 2013; Ungar-Sargon, 2015). Here, however, we focus exclusively on the empirical aspects of circumcision as they pertain to a published draft of the first-ever U.S. government policy on the subject, released in December of 2014. Our central aim is to analyse the contention of the U.S. Centers for Disease Control and Prevention (CDC) that the prophylactic benefits of non-therapeutic male circumcision, as carried out prior to an age of consent, outweigh its associated risks

in developed settings such as the United States, in light of the available evidence pertaining to this question.

Benefits versus risks

Do the benefits of circumcision outweigh the risks?² In recently proposed recommendations and an accompanying background report, an anonymous CDC working group has suggested that they do (2014a, 2014b). A similar claim was made in 2012 by an eight-member American Academy of Pediatrics (AAP) task force (2012a, 2012b), despite contrary policies from British, mainland European, and Australasian medical associations, all of which remain in force (e.g. British Medical Association, 2006; Royal Australasian College of Physicians, 2010; Royal Dutch Medical Association, 2010; see also National Health Service of England, 2016). In addition, the Canadian Paediatric Society (CPS), which has historically endorsed the AAP position, instead explicitly rejected it in its most recent policy (Sorokan, Finlay, & Jefferies, 2015).

What could explain this lack of international agreement with the U.S. view? There are several possibilities. At least one concerns the fact that 'The true incidence of complications after newborn circumcision is unknown', as acknowledged by the AAP task force (2012b, p. e772). But since 'complications' are one of the foremost risks of circumcision, and since their 'true incidence' has not been firmly established (see below), it becomes difficult to see how the benefits of the surgery could logically be asserted to outweigh them (Darby, 2015). This is especially the case given that, on the other side of the scale, the likelihood as well as the magnitude of the purported benefits of circumcision themselves are in dispute.

A further possibility has to do with ongoing disagreements over how to define 'complications' in the first place (see AAP, 2012a), as well as with differences of opinion concerning the relative weight or value to assign to individual benefits and risks. As AAP task force member Dr Andrew L. Freedman stated in a recent editorial, in addition to having 'insufficient information about the actual incidence and burden of nonacute complications', the AAP's 2012 assessment of benefits versus risks also suffered due to the 'lack of a universally accepted metric to accurately measure or balance the risks and benefits' (Freedman, 2016, p. 1).

To see the significance of this problem, which applies equally to the 2014 analysis by the CDC, consider the example of a modest reduction in the absolute risk of contracting certain sexually transmitted infections, a health benefit that is frequently attributed to – primarily adult – circumcision (see below). Considering that (1) children are not at risk of contracting sexually transmitted infections prior to sexual debut (an event that typically comes after the development of a capacity to provide informed consent to self-affecting interventions); (2) there are alternative modes of prevention that are less invasive, as well as less risky and more effective, than circumcision (i.e. safe sex practices when one does become sexually active); and (3) many of these infections can be treated effectively if they do occur, what is the weight or value that one should assign to this particular benefit?

For example, should it be considered 'worth' the risk, however slight, of a surgical mishap that causes permanent damage to the penile glans? Is it 'worth' the loss of the penile prepuce itself, which is a 100% risk of circumcision? Is it 'worth' the risk of

removing too much penile skin (i.e. more than was intended), leading to painful erections later in life (see e.g. van Duyn & Warr, 1962; Krill, Palmer, & Palmer, 2011; Thorup, Thorup, & Ifaoui, 2013)? The answer to these questions cannot be 'objectively' determined (see Johnsdotter, 2013). Instead, they will depend upon such factors as how much value one places on having intact versus modified genitalia, how willing one is to engage in safe sex practices (even if one is circumcised), and how much risk one feels comfortable taking on when it comes to a surgery performed on a physically and symbolically sensitive part of one's body (Adams & Moyer, 2015, p. 723; see also Earp, 2016).³

Finally, it has been argued that the appropriate counterbalance to the potential benefits of circumcision is not only the risk of surgical complications (whatever those turn out to be), but also its short-term, intermediate, and long-term adverse consequences, both physical and psychological (Darby, 2015). Due to a lack of adequate research into these questions, however, the entire spectrum of potential circumcision harms (i.e. surgical risks plus additional negative consequences) has never been fully described. Moreover, at least some of these potential harms are likely to be subjective in nature (e.g. feelings of loss or resentment; see Darby & Cox, 2009; Goldman, 1999; Hammond, 1999), and therefore highly variable across individuals, as well as difficult to quantify in a meaningful way (Darby, 2015; Darby & Cox, 2009; Johnsdotter, 2013).

The contention of the CDC

Notwithstanding all of the concerns just mentioned, in support of its contention that the benefits of circumcision do in fact outweigh the risks, the CDC working group cites a review article first-authored by a retired molecular geneticist named Brian J. Morris (Morris, Bailis, & Wiswell, 2014a). As noted previously in this journal, Morris is 'a long-standing advocate of mass circumcision' (Bell, 2015, p. 558), who now serves as the primary spokesperson for a pro-circumcision lobbying organisation called the Circumcision Academy of Australia (CAA). According to this group's website (http:// www.circumcisionaustralia.org), the CAA 'is not aligned with any medical body', but is rather a political entity seeking to expand health insurance coverage for non-medical circumcisions in Australia.4

Whether the CDC working group was aware of this context is unclear. Nevertheless, it approvingly quotes the benefit-to-risk ratio reported by Morris et al. (2014a) in its background report, which is stated as 100:1 in favour of circumcision. However, this is a figure that no other scientist, nor any recognised medical body to our knowledge, has ever been able to reproduce. Instead, it overstates the relative magnitude of the benefits of circumcision by a factor of approximately 100 according to the recent non-partisan analysis by the CPS (Sorokan et al., 2015). As the CPS states, 'the medical risk:benefit ratio of routine newborn male circumcision is closely balanced when current research is reviewed' (p. 4).

Given the prima facie implausibility of a 100:1 benefit-to-risk ratio in favour of circumcision, one might expect that the CDC working group would have sought out available criticisms of the paper by Morris et al. (2014a) in order to incorporate alternative perspectives. If so, they would have come across a critique published subsequently in the same journal, which states that the paper is 'marred by bias' and that its authors 'did not provide a dispassionate review' of the available evidence (Jenkins, 2014, p. 1588; but see Morris, Bailis, & Wiswell, 2014b). In a similar vein, Dr Kevin Pringle, Professor of Paediatrics and Head of Obstetrics and Gynaecology at the University of Otago, stated:

I find this paper to be extremely worrying. The most worrisome aspect is the emphasis on possible diseases that are reported to be significantly more common in the uncircumcised population and the complete lack of any attempt to accurately document the risk of the complications of circumcision. (2014)

Professor Pringle went on to state:

As I read their table, the authors suggest that performing 10 circumcisions will prevent 1 case of phimosis and/or 1 case of balanitis. [But] 80% of children with phimosis respond to the simple application of a steroid cream and only a small percentage need a circumcision. Similarly, many cases of balanitis are associated with phimosis and if the phimosis is treated, they have no further trouble. Paediatric surgeons see most of the cases of phimosis and balanitis. They are nowhere near as common as is suggested in this paper. (2014)

A new population-based study from Denmark, where non-therapeutic childhood circumcision is rare, lends support to Professor Pringle's impression. In a stable Copenhagen population with on average 10,858 boys born each year between 1996 and 2014, only 53 boys needed a circumcision for medical reasons before age 18 years during the calendar year 2014. This indicates that approximately 99.5% of Danish boys will go through infancy, childhood, and adolescence without any medical need to be circumcised (Sneppen & Thorup, 2016).

Finally, other authors have described a separate review by Morris and a co-author as being of 'low quality' on account of the 'high risk of bias ... introduced by the authors' well documented, unconditional support of the practice of circumcision' (Bossio, Pukall, & Steele, 2015, p. 1306; see also Donovan, 1999). In light of these considerations, it is not readily apparent why the CDC working group chose to rely so uncritically on the claims of Morris et al. in support of its main conclusion. For an Australian perspective, the working group might instead have cited the official 2010 policy of the Royal Australasian College of Physicians, which was recently re-affirmed after an evaluation of the AAP findings from 2012 (see Pringle, 2014; see also Forbes, 2015). This policy states: 'the frequency of diseases modifiable by circumcision, the level of protection offered by circumcision and the complication rates of circumcision do not warrant routine infant circumcision in Australia and New Zealand' (RACP, 2010, p. 5).

The value of an intact sexual organ

In a previous section, we noted that the appropriate counterbalance to the benefits of circumcision is not only the (unknown) risk of surgical complications, but rather the sum total of negative consequences, that is, the harms. One harm of circumcision that has been argued to be intrinsic to the procedure is the loss of healthy, functional tissue (Hutson, 2004). Yet in its background material, the CDC does not describe the known anatomy of the penile prepuce, nor does it discuss in detail the protective and sexual functions that have been attributed to it in the medical literature (2014b). One possible consequence of these omissions is that they may be taken to imply that the foreskin itself should be assigned a value of 'zero' in harm-benefit calculations. Yet such a valuation is uncommon outside of circumcising societies, and is inconsistent with normative medical evaluations regarding other functional parts of the body (Hutson, 2004).

The foreskin is a complex, double-layered structure, which protects the penile glans from environmental irritation, such as rubbing against diapers and clothing (Berry & Cross, 1956; Cold & Taylor, 1999). This elastic, motile sleeve of tissue has recently been shown to be the most touch-sensitive part of the penis (Bossio, Pukall, & Steele, 2016; consistent with Sorrells et al., 2007), composed of a moist mucous membrane on the inside and a protective skin layer on the outside. It is rich in specialised nerve endings and sensory structures involved in the normal functionality of the penis (Cold & Taylor, 1999), and it comprises up to 100 square centimeters in adult men, with reported mean values between 30 and 50 square centimeters (Kigozi et al., 2009; Werker, Terng, & Kon, 1998). While the scientific literature on the 'average' sexual consequences of circumcision is inconclusive and contradictory (Bossio, Pukall, & Steele, 2014, 2015; Johnsdotter, 2013) - and granting that circumcision is likely to affect different men differently, even when it is properly performed – at least two outcomes can be known with certainty due to the inherent nature of the procedure: first, any sensation in the foreskin itself is necessarily eliminated; and, second, any sexual (e.g. masturbatory) functions that require its manipulation are also of necessity precluded (Earp, 2015a, 2016).

Scientific inference from African trials

In its background report (CDC, 2014b), the CDC working group, like the AAP before it (2012b), emphasises three African trials concerning adult circumcision. These trials reported a modest reduction in the absolute risk of heterosexual, female-to-male human immunodeficiency virus (HIV) transmission (from 2.49% to 1.18%) during the first few years after circumcision in high-risk areas with a low baseline circumcision prevalence (CDC, 2014b). A number of concerns regarding both the internal and external validity of findings from these African trials have been raised (e.g. Boyle & Hill, 2011; Garenne, Giami, & Perrey, 2013; Green et al., 2010; but see Wamai et al., 2012 and further commentary by Bell, 2015), but even assuming that the reported efficacy estimates of the three African trials are scientifically valid, this does not necessarily imply that their findings are relevant elsewhere in the world.

The CDC does superficially acknowledge some of the problems associated with making scientific inferences from studies of adult males in Africa to newborn boys in the United States (2014b). However, the primary implication of such important caveats, namely that findings from the one context cannot responsibly be extrapolated to the other without a great deal of further investigation, is ultimately ignored. For example, the CDC background report (2014b) highlights two analyses of the costs and health consequences of circumcision which largely assume, based on unrepresentative observational data, that the findings from adults in Africa will apply directly to infant boys in the United States (Kacker, Frick, Gaydos, & Tobian, 2012; Sansom et al., 2010).

The inference is stretched even further. According to the CDC, all genitally intact adolescent U.S. boys and heterosexually active adult males (not only those in a few high-risk subgroups) 'should be informed about the significant, but partial, efficacy of male circumcision in reducing the risk of acquiring HIV and some STIs through heterosexual sex' (CDC, 2014a, p. 3). Since the absolute risk of acquiring HIV for the average intact heterosexual male in the United States is extremely low, however (McQuillan et al., 2006), the recommendation that all should be counseled about the 'significant' benefits of circumcision is arguably out of proportion (for an accessible discussion, see Bundick, 2009).

European experience

The CDC does not consider the situation in Europe, where rates of HIV are similar to or lower than those in the United States. In 2011, for instance, according to the Central Intelligence Agency (CIA) World Factbook, the prevalence of HIV/AIDS in the age group 15-49 years was several times higher in the United States (0.6%), where around 80% of men are circumcised (CDC, 2014b), than in several countries of northern and western Europe (0.1-0.3%), where circumcision is uncommon (CIA, 2011). While numerous variables undoubtedly play into these differences, the fraction of circumcised men in a given population is unlikely to be a determining factor.

Against this view, the CDC cites a modelling study by Kacker et al. (2012) suggesting that if the rate of male circumcision were to drop to around 10% as in Europe, then the United States would be destined to experience a significant increase in HIV infections and a heavy burden of associated healthcare costs (2014b). Unfortunately, this analysis, and another one by Sansom et al. (2010), relied on highly speculative trans-cultural and age-range extrapolations, and both models used estimates of complication rates that are unrealistically low (0.4% and 0%, respectively), as we will demonstrate below. Taken together, then, the cited cost-effectiveness estimates of newborn circumcision in reducing HIV in the United States (Kacker et al., 2012; Sansom et al., 2010) are overly optimistic, notwithstanding the fact that one of these estimates (Sansom et al., 2010) actually failed to show a cost-saving impact among the majority population of white males.

Procedural and post-operative pain in infants

It is now well established that infants and small children have the full capacity to experience pain, and that early life exposure to noxious stimuli may have long-lasting implications for the developing nervous system (Fitzgerald & Beggs, 2001). Consistent with this view, a different AAP task force that was formed to investigate infant pain (rather than circumcision) has recently concluded that:

Pain that newborns experience from routine medical procedures can be significant... Research suggests that repeated exposure to pain early in life can create changes in brain development and the body's stress response systems that can last into childhood. Because of this, a new American Academy of Pediatrics policy statement recommends [that] every health facility caring for newborns should use strategies to minimize the number of painful procedures performed. (2016)

Notwithstanding the clear importance of this issue, the problem of procedural and postoperative pain received only scant attention in the CDC's background report (2014b). The only cited study dealing with this question assessed pain scores among 583 boys ages 1-35 days who received local anesthesia (ring block) before they were circumcised (Banieghbal, 2009). Apparently, the CDC working group did not read the study carefully, but relied solely on the article's abstract, which states that 'only 6.5% of neonates under 1 week of age' experienced circumcisions with pain levels above a pre-defined threshold (Banieghbal, 2009, p. 359).

The quoted figure appears nowhere apart from the abstract (Banieghbal, 2009). An estimate of 6.7% is provided in a table, but that figure is miscalculated and should be 7.1%. While inaccurate reporting of key statistics is always regrettable, it is particularly worrying that the CDC would rely exclusively on such a report to minimise concerns about pain in infants.

In addition to this concern, the CDC missed entirely another key finding in the Banieghbal study. The ring block injections administered to all the boys resulted in above-threshold pain in fully 31% of <1 week-old boys and 85% of 1-5 week-old boys. In other words, simply being administered the anesthesia proved painful for many infants to a clinically significant degree. Additionally, among 1-5 week-old boys, 71% experienced above-threshold pain during the subsequent circumcision (Banieghbal, 2009). Consequently, the CDC's statement that '93.5% of neonates circumcised in the first week of life with appropriate analgesia gave no indication of pain on an objective, standardised neonatal pain rating system' (2014b, p. 25) misrepresents the overall findings from the study.

Finally, pain at the operating table is not the only pain to factor in. In one study of postoperative discomfort in 57 neonatally circumcised boys, parental assessment revealed that 16% of the boys experienced moderate to severe post-operative discomfort, while the remaining 84% were judged by their parents to have mild discomfort (Smith & Smith, 2000). In another study of 710 newborn boys in Michigan, 71% of parents reported varying degrees of circumcision-related pain in their infants (ranging from "minimal" to "much more than acceptable") up to six weeks after the surgery (Freeman et al., 2014).

Complication rates after circumcision

In a section evaluating the safety and risks associated with circumcision (CDC, 2014b), the CDC focuses on immediate surgical risks and short-term post-operative complications, as identified in U.S. register data. Three studies published between 16 and 40 years ago are given particular attention (Christakis et al., 2000; Gee & Ansell, 1976; Wiswell & Geschke, 1989). However, each of these studies was designed to address complications occurring no later than one month after neonatal circumcision; indeed, two studies dealt exclusively with complications occurring just during the post-partum hospital stay (Christakis et al., 2000; Gee & Ansell, 1976). As pointed out by others, basing evaluations of the frequency of complications after infant circumcision on insufficiently detailed and incomplete register data will likely produce 'a significant underestimation of the true frequency of complications' (Pieretti, Goldstein, & Pieretti-Vanmarcke, 2010, p. 517). For instance, serious or even fatal complications from post-operative bleeding or infection due to circumcision may plausibly be recorded as hemorrhage, circulatory collapse, or septicaemia without mention of the surgery that caused it (Dr A. Rotta, personal communication, 11 April 2016). Likewise, when complications are treated in other hospitals or clinics than those responsible for the original circumcision, the general inability to cross-link register data in countries without a unique personal identifier for every citizen (such as the United States), may preclude the identification of such post-operative complications as being truly circumcision related (see Weiss, Larke, Halperin, & Schenker, $2010).^{7}$

In addition, a recent Canadian study by DeMaria, Abdulla, Pemberton, Raees, and Braga (2013) found:

Most physicians performing neonatal circumcisions in our community have received informal and unstructured training. This lack of formal instruction may explain the complications and unsatisfactory results witnessed in our pediatric urology practice. Many practitioners are not aware of the contraindications to neonatal circumcision and most non-surgeons perform the procedure without being able to handle common post-surgical complications. (p. 260)

Such a lack of formal training in circumcision may also prevent some doctors from recognising certain complications in the first place; complications that are not recognised cannot be reported.

Particularly worrying is the CDC's assessment of meatal stenosis, a narrowing of the urethral opening which is seen at far higher rates in circumcised boys compared to genitally intact boys (Bazmamoun, Ghorbanpour, & Mousavi-Bahar, 2008; Berry & Cross, 1956; van Howe, 2006; Joudi, Fathi, & Hiradfar, 2011). In support of its contention that complication rates of circumcision are trivially low, the CDC cites a study by El Bcheraoui et al. (2014). These authors looked at hospital billing claims to evaluate the broader issue of urethral strictures (which includes meatal stenosis) during the first 180 days after surgery. However, a narrowing of the urethral meatus may take considerably longer than 180 days for parents or doctors to detect (Berry & Cross, 1956; Campbell, 1943; Joudi et al., 2011; Upadhyay, Hammodat, & Pease, 1998), which means that the chosen window of analysis was too short. Accordingly, the observed low frequency of urethral strictures (0.01%) – and therefore the low overall frequency of complications for boys circumcised in infancy (0.4%) (El Bcheraoui et al., 2014) – most likely reflects a combination of insensitive research data and insufficient follow-up.

Given such limitations affecting register-based studies, more realistic estimates of complication rates should be drawn from clinical studies. However, surprisingly limited data exist to estimate the true rate of late complications occurring months to years after nontherapeutic circumcision. In a 2010 review of complications after newborn, infant, and childhood circumcisions, where no clear and consistent distinction was drawn between (1) ritual circumcision of healthy, predominantly infant boys and (2) therapeutic circumcision of predominantly older boys with foreskin pathology, investigators identified 52 studies from around the world, of which only one small prospective study from 1988 (n = 43; mean age at circumcision = 6.5 years) provided information about adverse outcomes beyond the first year after the surgery (Weiss et al., 2010). Indeed, the only adverse outcome for which a reasonable number of reports provide frequency estimates based on direct clinical examination is meatal stenosis, which is broadly considered to be one of the most common complications after circumcision (Bazmamoun et al., 2008; van Howe, 2006; Joudi et al., 2011; Kajbafzadeh et al., 2011; Mondal et al., 2013; Patel, 1966; Persad, Sharma, McTavish, Imber, & Mouriquand, 1995). From such clinical studies, it appears that between 5% and 20% of boys undergoing non-therapeutic circumcision by means of conventional surgical techniques will develop meatal stenosis.

In one Iranian study, which was published only as a meeting abstract, 5% of 102 neonates circumcised by the PlastiBell method developed meatal stenosis during on average 9 years of follow-up; the corresponding proportion was 15% of 105 neonates circumcised by means of classical circumcision surgery preceded by ligation of the frenular artery (Kajbafzadeh et al., 2011). Similarly, a study from the United States reported a 7% prevalence of meatal stenosis in 1009 neonatally circumcised boys older than 3 years (van Howe, 2006). The CDC downplayed this finding (2014b), however, preferring instead a report from

Iran, which described a frequency of only 0.9% for meatal stenosis in boys ages 6-12 years (Yegane et al., 2006). Yet this is flawed referencing: the reported rate of meatal stenosis in these Iranian boys was for all circumcised boys, most of whom (97.18%) were circumcised after the neonatal period.

In several other clinical studies relying on direct penile inspection from as varied geographical areas as the United States, the United Kingdom, Bangladesh, and Iran, proportions of boys developing meatal stenosis after conventional circumcision surgery in infancy or early childhood were 7% (Bazmamoun et al., 2008), 8% (Patel, 1966; Persad et al., 1995), 10% (Mondal et al., 2013), or 20% (Joudi et al., 2011). All of these studies were ignored by the CDC. And yet their estimates are more consistent with the 7% meatal stenosis rate reported in the U.S. study by van Howe (2006), which the CDC dismissed, than with the 700-fold lower 0.01% urethral stricture rate reported by El Bcheraoui et al. (2014), to which the CDC paid particular attention.⁸

While no study is free of limitations, reported rates of 5–20% for post-circumcision meatal stenosis in clinical studies (Bazmamoun et al., 2008; van Howe, 2006; Joudi et al., 2011; Kajbafzadeh et al., 2011; Mondal et al., 2013; Patel, 1966; Persad et al., 1995) call attention to the need for extra caution in interpreting reports of absolute complication rates based on register data, such as that relied upon by El Bcheraoui et al. (2014).

Other studies confirm the impression that the CDC working group may have seriously underestimated the burden of complications after newborn circumcision. For instance, Pieretti et al. (2010) reported from a hospital department of pediatric surgery in Boston (just one of several referral centers in the region; total number of circumcisions unknown) that of the 8967 children (both boys and girls) operated on between 2003 and 2007, 4.7% of the operations were for late complications occurring 2-49 months after neonatal circumcision. Another indication of the magnitude of the problem was that, of all outpatient visits (again, of both boys and girls) to the department of pediatric urology at the same hospital during a one-year period between 2007 and 2008, 7.4% were for complaints related to newborn circumcision (Pieretti et al., 2010).

Summary and conclusion

In this brief analysis, we have identified numerous scientific and conceptual shortcomings in the 2012 circumcision policy from the AAP, as well as the more recent draft guidelines issued by the CDC. With respect to the latter, these included: (1) failure to provide a thorough description of the normal anatomy and functions of the penile structure being removed at circumcision (i.e. the foreskin); (2) failure to consider the intrinsic value to some men of having an unmodified genital organ; (3) undue reliance on findings from sub-Saharan Africa concerning circumcision of adult males (as opposed to infants or children); (4) uncritical reliance on a prima facie implausible benefit-risk analysis performed by a self-described circumcision advocate (see Davey et al., in press); (5) reliance on misreported statistics to downplay the problem of pain in the youngest of boys; (6) reliance on incomplete register data to assess the frequency of short-term post-operative complications associated with circumcision, leading to a likely underestimation of their true frequency; and (7) serious underestimation of the late-occurring harms of circumcision presenting months to years after the operation (most notably meatal stenosis). In light of these considerations, we believe that the CDC's overall assessment of benefits versus harms ('risks') of the surgery should be interpreted with extraordinary caution.

The apparent underassessment of meatal stenosis alone, possibly the most common late-occurring complication after neonatal circumcision, means that the CDC's claim of markedly lower complication rates in boys circumcised as infants ('less than ½ per cent'), compared to boys circumcised at ages 1–9 years ('approximately 9%') and those circumcised at age 10 years and older ('approximately 5%') (2014a, 2014b), is almost certainly inaccurate.

Indeed, with reported rates of meatal stenosis in neonatally circumcised boys in the 5–20% range, and with estimates of clinically significant procedural pain in around 30% (and some level of post-operative pain and discomfort in most, if not all newborn boys undergoing the operation), the least problematic age to circumcise a boy (if at all), even from a purely medical standpoint, may well be when he is old enough to decide for himself. This conclusion is supported by the recent evidence showing that very few genitally intact boys – 0.5% according to the new population-based study in Denmark (Sneppen & Thorup, 2016) – will need a circumcision for medical reasons before the age of 18.

Notes

- 1. It is important to emphasise that this draft was made public, garnering much coverage in the popular media, prior to having been subjected to a formal peer review. In a notable turn of events, at least one medical professional who was invited by the CDC to perform such a review subsequently made his comments available online (van Howe, 2015). This freely accessible, 200+ page review is strongly critical of the CDC research and analysis, as are several other expert commentaries that were posted to the CDC website in response to a call for public feedback (e.g. Kupferschmid et al., 2015). Nevertheless, it is currently unclear whether, or when, the CDC intends to revise its recommendations in light of such critical feedback or else formalise them (or a modified version of them) as official policy.
- 2. It is widely acknowledged that removing the foreskin of the penis may confer certain health benefits, in much the same way that removing healthy tissue from any other part of the body could be expected to reduce the risk of medical problems affecting or introduced via that tissue (see e.g. Kluge, 1994). For example, routinely removing one testicle from every male child would almost certainly reduce the individual boy's risk and the population-wide incidence of testicular cancer; but the costs, harms, and other disadvantages (sometimes misleadingly referred to as 'risks', see Darby, 2015) of prophylactic testicle removal would need to be factored into the equation, along with an array of moral considerations concerning autonomy, consent, and bodily integrity (not addressed in the present paper). In the case of male circumcision, then, the question is not whether certain health benefits may in fact ensue from the sheer surgical removal of the foreskin, but whether, in light of alternative, less invasive, means of achieving the same desired health outcomes, the benefits are sufficient to outweigh the costs, harms, and other disadvantages (i.e. 'risks'), some of which may be subjective in nature and therefore difficult to quantify (see e.g. Adams & Moyer, 2015; Darby & Cox, 2009; Johnsdotter, 2013).
- Moreover, as AAP task force member Freedman (2016) has additionally noted, in the United States, at least,

although parents may use the conflicting medical literature to buttress their own beliefs and desires, for the most part parents choose what they want for a wide variety of non-medical reasons. There can be no doubt that religion, culture, aesthetic preference, familial identity, and personal experience all factor into their decision. (p. 1)

Consistent with this perspective, Freedman has stated in a separate interview:

I circumcised [my own son] myself on my parents' kitchen table on the eighth day of his life. But I did it for religious, not medical reasons. I did it because I had 3,000 years of ancestors looking over my shoulder. (see http://www.thejewishweek.com/features/ new-york-minute/fleshing-out-change-circumcision#zpW7AFLaLTQsqJMH.99)

Granting these considerations, a further question is raised, however, regarding who should get to decide about circumcision in light of such non-medical factors (about which there is no universal agreement about how to weigh them): the parents, as assumed by Dr Freedman (including in his own case), or the individual who must undergo the surgery and therefore live with its consequences, positive or negative, as judged in light of his own considered preferences and values (i.e. the child himself, when he reaches an age of competence)? As we have just seen, even purely 'medical' factors may be weighed differently in the minds of different individuals; how much more contentious might non-medical factors be?

- 4. It appears that such expansion of coverage would be of direct financial benefit to several CAA board members, including the organisation's inaugural president, Dr C. Terry Russell (Russell Medical Center, 'Trusted Australian Circumcision Since 1993', http://www. circumcision.net.au), along with board members Dr Mojtaba Athari (Melbourne Vasectomy & Circumcision, http://www.mvandc.com.au/circumcision/), Dr Luke Bukallil (who has performed 'over 2000' circumcisions, http://drluke.biz/Circumcision/circumcision.htm), Dr Mohamed Hajoona (Victoria Circumcision Clinic, http://www.vcc.net.au), Dr Colin C.M. Moore (The Australian Center for Cosmetic and Penile Surgery, http://www.drcolinmoore. com), and Dr Anthony Dilley (who 'conducts up to 40 circumcisions per week', http:// www.dailytelegraph.com.au/news/nsw/unkindest-cut-of-all-back-in-favour/story-e6freuzi-1225991948585). The group's political petition to 'restore elective male circumcision to public hospitals ... increase the Medicare rebate [and] ensure [that] Medicare applies to prophylactic circumcision not just circumcision for medical problems' can be found here: http:// chn.ge/1otJ9Bv. A similar petition posted separately by Brian Morris is available here: https:// www.change.org/p/nsw-ministry-of-health-restore-elective-male-circumcision-to-publichospitals.
- 5. Please note that the reply to Jenkins by Morris et al. (2014b) suffers from many of the same problems that Jenkins pointed out in his critique of their original paper. For example, most of the references in the reply are to other contested papers by Morris and colleagues (with no mention of published criticisms of those papers), creating what others have described as a 'rabbit hole' of selective self-citation (Svoboda & van Howe, 2013).
- 6. Alexandre T. Rotta, M.D., FCCM, is the Linsalata Family Chair in Pediatric Critical Care and Emergency Medicine, and Chief, Division of Pediatric Critical Care, Rainbow Babies & Children's Hospital (University Hospitals of Cleveland); as well as Professor, Department of Pediatrics, Case Western Reserve University School of Medicine. Dr Rotta describes some of the difficulties in linking circumcision-related complications with the circumcision surgery itself due to coding issues in an interview that is available online at the following link: https://www. youtube.com/watch?v=x_BohYj-VMw.
- 7. Further, indirect support for this view comes from a recent review paper by Springer (2014) concerning complications associated with surgery for hypospadias, another penile surgery that is often carried out in infancy or early childhood. As summarised by Carmack, Notini, and Earp (in press), on the issue of underreporting, Springer 'identified numerous barriers to complete reporting of surgical, functional, cosmetic, and quality-of-life outcomes' associated with the surgery, including

numerous techniques in use; most publications reflecting single-center and singlesurgeon retrospective case series with limited follow-up periods and small numbers of patients undergoing follow-up; transition of care from pediatric to adult specialists, thus limiting follow-up into the period when sexual activity is likely to occur;



assessment of cosmetic outcomes by surgeons who may be biased to approve of their own 'work' ... and difficulty assessing sexual function and behavior in young adults. (p. 6)

Many of these 'barriers to complete reporting' of adverse outcomes are likely to apply to circumcision as well, although the matter has not been adequately studied to date. This lack of direct and comprehensive research into the issue calls for still further caution in interpreting available estimates of circumcision-related complications.

8. Indeed, the CDC working group mistakenly listed the El Bcheraoui et al. (2014) paper twice – as references 158 and 164 – in its background report (CDC, 2014b), and as one of only 15 highlighted references in the shorter recommendation document (CDC, 2014a).

Disclosure statement

Both authors have published articles and taken part in national and international debates concerning health-related, sexual, and ethical aspects of male and female circumcision. From a medical ethics perspective, they are generally sceptical about the advisability of performing non-therapeutic genital surgeries on minor children.

Funding

The authors report no financial or personal relationships with other people or organisations that could inappropriately influence their work.

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