Does circumcision cause SIDS?
A point-by-point response to Eran Elhaik's study

Medical headlines in July promoted a study that indicated that infant circumcision may increase the risk of sudden infant death syndrome (SID), also known as cot teadh. Dr. Eran Elhaik of the University of Sheffield in Great Britain authored the study. In 2016 Elhaik proposed a hypothesis that stressful events during infancy, including circumcision, increased a risk of SIDS.

One must be skeptical when activists on one side of a partisan issue report study results that support their agenda. Skepticism is especially important in the circumcision debate. A 2007 study funded by an anti-circumcision group found that circumcision removes the most sensitive parts of the penis. And a 2011 study coauthored by the Intact America strategy advisor found that circumcised men have a 4½ times greater risk of erectile dysfunction than uncircumcised men. Although both studies had potential methodological flaws and the authors advised caution, intactivists have seized on the results to "prove" that circumcision causes harm.

This paper presents a point-by-point response to the present study. While the study also addressed the possible effects of premature birth, this response focuses solely on the portions that are about newborn circumcision.

The text of the study is in **black**, and my responses are in **blue**. The study can be accessed at this link:

https://www.biorxiv.org/content/biorxiv/early/2018/06/07/339465.full.pdf

Introduction

> To test the predictions of the allostatic load hypothesis for SIDS, we identified two common stressors [5], male neonatal circumcision (MNC) and premature birth, for which latitudinal data were available and tested their association with SIDS. Both stressor are male-biased [11] and may explain the male predominance of SIDS, whereas the first stressor may also explain the lower SIDS rates in Hispanics. MNC is associated with intraoperative and postoperative risks including bleeding, shock, sepsis, circulatory shock, and hemorrhage [12-14]

The American Academy of Pediatrics reports that "acute complications are usually minor and most commonly involve bleeding, infection, or an imperfect amount of tissue removed." Circulatory shock and sepsis are extremely rare complications. This author is unaware of a case of neurogenic shock as a complication of circumcision.

Weiss [12] examined 16 studies performed in 12 countries, involving a total of 26,645 patients. Only 4 of the studies, involving 1,410 patients (5%), were conducted in the countries that Elhaik used here. So it's questionable whether all of the risks identified in the Weiss study are relevant to the countries reviewed in this study. Neverthless, Weiss reported that the most common complications … were minor including bleeding (9%) and meatal stenosis (3.5%)." Weiss reported just one case of hemorrhage, but no cases of shock or sepsis. This author wrote an extensive refutation of Boyle [13], analyzing every complication. Boyle did not report any cases of circulatory shock or sepsis. Boyle claimed that "some infants do not cry because they go into shock." To support this claim, Boyle cited Svoboda, who provided no evidence for the assertion. Edler [14] reported 4 cases of circulatory shock out of more than 140,000 infant circumcisions.

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> that can result in death [14, 15].

Cases in which circumcision is a direct cause of death (e.g. via excessive bleeding) are not relevant to the issue at hand, which is whether babies who have been circumcised are at a greater risk for sudden infant death syndrome. A baby who bled to death cannot be said to have died of SIDS.

At any rate, Edler [14] reported one death (in 2012, from severe blood loss) in 20 years of circumcision in Sweden, Denmark, and Norway. More than 7,000 circumcisions are performed in Scandanavia every year, or 140,000 over the 20-year period. Blackwell [15] reported on the 2013 death of a 3-week old infant in Canada. By comparison, Speert reported 1 death in 566,000 circumcisions in New York during 1939-1951. Wiswell reported no deaths in 100,000 circumcisions performed in U.S. Army hospitals during 1980-1985. Death caused by circumcision of a healthy American newborn is extremely rare.

> MNC may also cause severe and long-lasting pain, trauma, and psychological impairment due to the circumcision procedure that involves maternal separation, restraint to a board, and the removal of sensitive penile tissues that contain numerous nerve endings [16-21].

Page [16] merely cited Taddio [21]. Fleiss [17] wrote in a 1997 article in *Mothering* magazine that "circumcision cuts off … more than 20,000 nerve endings." He cited a 1932 paper in which a researcher had counted the number of nerve endings in a specimen that measured one square centimeter. However Fleiss made several dubious assumptions in order to arrive at that figure. Goldman [18] cited several books and studies, none of which discussed trauma that occurs during the neonatal period. Despite admitting that "there is no empirical research on circumcision trauma and memory," Goldman speculated that newborns *might* be able to retain memory of trauma. However Strange reported that "early memories are extremely rare" and found that adults "appeared to have a reduced threshold for accepting" details regarding [false] memories from age 2. Strange concluded that "childhood amnesia increases [the] susceptibility to false suggestion." Hama [19] studied the effect of injury in neonatal animals, not the effect of infant circumcision on adolescents and adults. Bear in mind that Fleiss, Goldman, and Hama were or are anti-circumcision activists who had a motive to "discover" harmful effects. Taddio [20] found that certain pain relief was effective in the "prevention of pain from circumcision in neonates." Taddio [21] tested only for the Gomco method and only for Emla as pain relief. Follow-up consisted of one measurement during a 4- or 6-month vaccination visit; measurements at older ages were not taken. None of these sources provided empirical evidence to support Elhaik's claim that a neonatal event can cause longlasting psychological trauma and impairment.

Elhaik implied that circumcision involves separation from parents and restraint on a board. But parents often stay with the child when the procedure is performed in a hospital or medical office. Some parents hold their son during the procedure. And a restraint board is never used for Jewish ritual circumcisions; typically a grandfather holds the boy.

> Since MNC preference is largely cultural, populations can be classified into Anglophone countries (high MNC rate) and non-Anglophone countries (medium to low MNC rate [22, 23]) (Table S1). If MNC is a risk factor for SIDS, SIDS rates would be higher in Anglophone countries, where MNC is highly prevalent [22], compared to nonAnglophone countries, which traditionally have opposed circumcision [23]. US populations also differ in their MNC practices. Between 2005 and 2010, non-Hispanic Whites were the largest group performing MNC (90.8%), followed by non-Hispanic Blacks (75.7%) and Mexican

Americans (44%). If MNC is a risk factor for SIDS, in addition to their low SIDS rates we can also expect Hispanic populations to exhibit lower male bias than non-Hispanics.

Data collection

Global male neonatal circumcision (MNC) rates per country (2005-2006) were obtained by searching for 'neonatal circumcision' and country in Google Scholar, Google, and PubMed. Similarly to [28], MNC rates for the remaining countries that could not be obtained through peer reviewed journals and whose adult circumcision rates were estimated by the WHO to be <20% [22] were estimated from the total percentage of Muslims [29] and Jews [30] in the country, as both populations were reported to have 100% circumcision rate [31].

A 100% Muslim rate is based on a false assumption that circumcision of Muslim boys must occur during infancy. Anwer reported that "Islamic traditions do not provide specific recommendations on the timing of the ritual; hence the age among Muslims varies widely." Anwer found that one third of the boys in a study in Pakistan were circumcised after 2 months, the average age was 1 year old, and the oldest was 13 years. Sahin reported a study in Turkey; the median age at the time of circumcision was 6 years, and less than 15% were circumcised before 1 year of age.

US statewise MNC rates (2009-2013) were obtained from [32].

We collected global SIDS prevalence data for 15 countries from 2004-2013 [9, 33, 34]. Year-matched MNC and SIDS data were available for Australia, Canada, New Zealand, and US. Global SIDS and MNC data are summarized in Table S1. All US SIDS data were obtained from the Centers for Disease Control and Prevention (CDC) Wonder [9]. US statewise MNC rate and male:female SIDS ratio are summarized in Table S2. US statewise male bias SIDS data (1000*MSIDS rate/FSIDS rate) between 1999 and 2016 are summarized in Table S3.

Results

SIDS prevalence varied greatly among the studied countries, ranging from 0.06 to 0.82 per 1,000 births ($x=0.38$, $\sigma=0.22$) (Figure 1). SIDS prevalence was the lowest in the Netherlands (0.06) and highest in the US (0.82) and New Zealand (0.8)

Elhaik failed to explain how his hypothesis accounts for New Zealand, which has an infant circumcision rate of perhaps 10% and the highest SIDS rate.

Data analyses

We argue that the practice of MNC can explain those differences and showed that large proportions of SIDS and SIDS variation between genders in the US can be explained by the MNC rates but not prematurity. Our results suggest that MNC contributes to the high mortality and gender-bias. That the equivalent practice of female genital mutilation is illegal in a growing number of countries [69] further increases that bias. In addition, females benefit from the protective effect of their sex hormones like estrogen against stressful and painful experience early in gestation [70-72]. We thereby surmise that the

> gender variation in SIDS is due to the dual legal-biological protection that females enjoy and that eliminating or postponing MNC may decimate the gender bias but not completely eradicate it.

Actually the relevant factor is that infants are not typically subjected to female genital mutilation. Irrespective of its legal status, the global rate of ***infant*** victims of FGM is practically zero.

> Our finding that MNC is associated with SIDS is not surprising. Circumcision is associated with intra operative and postoperative risks, including bleeding, shock, sepsis, circulatory shock, hemorrhage, pain, and long-term consequences [12-14, 73-76]

Weiss, Boyle, and Edler [12-14] have been addressed previously in this paper. Frisch [73] did not cite any case of shock or sepsis. Frisch claimed that a study reviewed by the AAP found 2 cases of hemorrhage for every 100 circumcisions. In fact the rate was for hemorrhage from *tonsillectomy*, not circumcision. Frisch admitted that he lacked conclusive evidence of "long-term psychological, sexual, and urological effects." Kaplan [74] cited Fredman regarding two cases of death from sepsis during a 10-year period. Fredman is not available online, so this avenue cannot be further explored. Kaplan did not cite any cases of shock, nor did he report on long-term effects. Kirkpatrick [75] discussed two cases of premature infants (4.1-4.4 lbs) who suffered life-threatening septicemia in 1972; fortunately both recovered. Mano reported that just 0.32% of infants were bleeding following ritual circumcision.

While bleeding is the most common complication, these citiations show that shock, sepsis, circulatory shcok, and hemorrhage as complictions of newborn circumcision are rare or non-existent. None of the citations provide proof of long-term consequences.

> – all of which contribute toward allostatic load [14, 15] and thereby SIDS through various mechanisms [5]. For instance, circumcision reduces the heart rate [20]

On the contrary, Taddio reported that circumcision *increases* the heart-rate. Taddio found that the application of lidocaine–prilocaine reduces the amount of the increase. She added that other "techniques are more effective [at decreasing pain] than applying lidocaine–prilocaine cream … and preventing large increases (of up to 60 beats per minute) in the heart rate."

> and together with the loss of blood there is a danger of reducing the blood volume, blood pressure, and the amount of oxygen reaching the tissues [5, 77]. A reduced blood pressure has been associated with obstructive sleep apnea (OSA), a condition where the walls of the throat relax and narrow during sleep, interrupting normal breathing [77, 78].

Ellhaik explained that "an infant has only 11 ounces of blood, and he may easily lose 1 to 2 ounces in circumcision, the equivalent of two to four blood donations for an adult." But normal blood loss during circumcision is less than 2 mililiters. A loss of 1-2 ounces (30-60 mililiters) would be enormous. Even so, a baby would have replaced the blood loss long before reaching the high-risk period for SIDS.

> Unsurprisingly, SIDS victims experienced significantly more frequent episodes of OSA [79]. Preterm neonates experience over twice the rate of bleeding complications than full-term neonates [80]. MNC-related complications are unavoidable [13, 14, 80-82]

Boyle [13] and Elder [14] were previously addressed in this paper. Litwiller [80] found a 12% rate of bleeding complications for circumcisions in which the Gomco method was used. Frisch [81] compared the complication rates in various studies. Park [82] examined men who visited a Korean medical facility for urological treatment. Park found that newborn circumcision was associated with a 0.30 inch shorter erect penile length. (Park noted that newborn circumcision "offers numerous health benefits and protections against certain medical conditions.") None of these sources provide evidence for Elhaik's claim that circumcision complications are inevitable.

> and in tandem with the lack of evidence of a meaningful and relevant health benefits to the infant,

The AAP reported that circumcision reduces the incidence of urinary tract infection during the first year. A lower risk of UTI is certainly a meaning and relevant health benefit to an infant. Moreover circumcision performed during infancy is associated with a reduced lifetime risk of penile cancer.

several countries chose to opt out of the operation [83].

The citation of the Canadian Paediatric Society position statement on newborn male circumcision [83] is quite remarkable, as the CPS position statement contradicted Elhaik's assertions. The CPS identified several "potential benefits," including phimosis treatment, UTI reduction, STI reduction, and cancer reduction. The CPS advised that parents should receive "the most up-to-date, unbiased and personalized medical information available so that they can weigh the specific risks and benefits of circumcising their son." The CPS reported Canada's infant circumcision rate at 32% - hardly an "opt out."

> For instance, in 1949, Gairdner's report [84] that 16 out of 100,000 UK boys under 1-year old died due to circumcision prompted the British government to exclude circumcision coverage from the National Health Service.

On the contrary, Darby asserted that the British decision was based on a lack of resources. With the UK in recovery from the ravages of World War II, "most physicians had real trouble justifying [the procedure] in the climate of near-poverty." The current British Medical Association position recognizes "that parents should be entitled to make choices about how best to promote their children's interests." The BMA admitted that "the medical evidence is inconclusive."

> Until the late 19th century, Jews were the only group practicing exclusively MNC in Europe [23]. It is thereby of interest to ask whether Jews were familiar with the association between MNC and SIDS.

Elhaik's statement about an association is premature. He even admitted that this study does not *prove* an association. He should execise caution and refer to "**a possible** association between MNC and SIDS."

> Elhaik [5] already showed that MNC was known to be a deadly practice for over a millennium and prompted the splintering of Reform Judaism from Orthodox Judaism in the nineteenth century. Here, we argue that several Jewish customs associated with MNC reflect the footmarks of SIDS, centuries before it was defined. Jewish ritualistic circumcision, as practiced today, emerged only during the second century AD [85]. It was also around that time that the myth of the baby-killer Lilith, a beautiful, taloned foot demoness [86], became prevalent [87]. Originally one of many Mesopotamian

> demons, Lilith clawed her way through the demonic hierarchy, extending her influence over time until she became Samael's (Satan) wife around the 13th century [86]. Deceiving Lilith into believing that the newborn was a girl by letting the boy's hair grow and even dressing him in girl clothes during infancy were the most effective means to avoid her harm. This Middle Age tradition [88] is still being among Orthodox and even secular Jews who avoid cutting a boys' hair for the first three years. Another ancient tradition is the "Night of Watching," a ceremony held on the night preceding circumcision to guard the newborn throughout the night against Lilith [89]. In some ceremonies the guests were particularly loud throughout the night to prevent the infant from succumbing to death. Overall, these practices are a testament to Jews' beliefs that 1) sudden death was and still is highly prevalent; 2) there exists a major male bias in these otherwise random infant deaths; 3) circumcision is associated with sudden deaths; and 4) sudden deaths occur at night – all of which are the hallmarks of SIDS.

It seems peculiar for a scientific paper to cite ancient mythology as evidence for a contemporary medical claim. At any rate, if Elhaik wants to consider the character of Lilith, he must consider the entire folklore. It would be disingenuous to pick and choose which parts of the character align with his hypothesis while ignoring parts that may conflict. Barkin-Kamil explained that Jewish boys "are especially vulnerable to her influence **until the ninth day after their birth**, and girls until the twentieth day." And Elhaik has acknowledged that "the first week of life [is] a time when a meager percentage of SIDS deaths occur." The vast majority would occur between one and four months, which is long past the danger posed by Lilith. Jewish mythology does not support an association between circumcision and SIDS.

> Unfortunately, there are limited data of the SIDS prevalence in Israel due to religious limitations on conducting autopsies [90]. Interestingly, Israeli health officials reported that, unlike in other countries, Israel saw no reduction in SIDS prevalence following the BTS campaign [91].

Siegel-Itzkovich [91] explained the unfortunate reason – medical professionals and parents in Israel were not following the updated guidelines for the prevention of SIDS.

> Our findings suggest that MNC, the most common unnecessary surgery in the world, is a major risk-factor for SIDS.

It would be helpful if Elhaik would explain how his assertion that infant circumcision is a major risk factor aligns with the position of New Zealand having the highest SIDS rate (0.80/1,000) and the position of Norway (0.30) similar than Canada (0.33). New Zealand and Norway have very low circumcision rates.

> Circumcised infants living in a stress-fraught environment, born prematurely, or have an existing genetic predisposition to sudden death would be at the highest risk of SIDS. While the risks of preterm births are well recognized, the debate concerning MNC is polarized between ethical concerns [99]

Referring to a polarized ethics debate, Elhaik provided only one side of the debate.

> and financial motives [100, 101]

Margulis [100] reported that foreskins are sold to bio-tech companies for treatments for burn victims, medical research, and cosmetics manufacturing. She claimed that hospitals charge as much as $20,000 to perform the procedure. She referred to an article on a health care website, but didn't explain that the story referred to the cost to circumcise a 14 month-old boy. Researching the price to circumcise a baby, author Rachel Zimmerman found that a hospital would charge roughly $2,800, while a local mohel quoted a rate of $1,400-$1,900 (which included $500 for anesthesia.) A 2011 *New York Times* article reported that the cost of a newborn hospital circumcision was $200-$400. Margulis didn't explain why physicians would have a financial motive to recommend *newborn* circumcision if they can charge from 8 to 100 times more when the patient is a year older. At any rate, a physician she interviewed scoffed at the idea of a financial motive. Dr. Asseem Shukla said, "As a pediatric urologist, it's the least-paying procedure that we do, but we do them as a service. I laugh when people think we do it for the money."

Hill [101], claimed that doctors would lose income and be exposed to financial liability "if societies were honest about the risks associated with circumcision and the certain loss of physiological function." His assertion presumes that a circumcised penis doesn't function properly. Hill provided no evidence for his accusation that doctors derive a significant income from performing elective circumcisions. And it's unlikely that a circumcision practitioner would face liability for performing a routine medical procedure that is presumed legal and is endorsed by the AAP, the United States government, and the state of California. The U.S. Constitution expressly prohibits ex post facto laws *(Article 1, Section 9, Clause 3 and Article 1, Section 10)*. It should be noted that Hill is vice president of a prominent anti-circumcision group and he holds a position of honor in the anti-circumcision movement for his three decades of activism.

> clouded by alleged medical benefits,

By sneering at well-documented medical benefits as merely *alleged* – while providing no such skepticism regarding *financial motives*, Elhaik made his anti-circumcision bias clear.

> with little awareness of the long-term risks for infants.

Elhaik provided no evidence for his assertion regarding long-term risks. The AAP identified "good and fair evidence that sexual function is not adversely affected in circumcised men compared with uncircumcised men." According to the WHO, medical studies show no evidence that circumcision affects sexual enjoyment and satisfaction.

> Although the conclusions of our ecological study should be verified in a cohort study with properly matched infants [102], some recommendation can be implemented immediately at little cost, such as: eliminating neonatal circumcisions when possible, postponing non-medical circumcisions to later ages,

Those recommendations do not follow based on the low quality and significant limitations of this study.

> informing parents of the risks in MNC, and applying pain management techniques to neonates that experience repetitive pain. MNC data should also be collected and tested in prospective SIDS studies.

Limitations

> This study has significant limitations, many of which are not due to the study design and are common to all SIDS studies. First, as in all ecological studies, correlation is not causation, and causation cannot be inferred from correlation alone.

And yet the author insisted that his study showed that circumcision is a major risk factor for SIDS. Anyone who offers this study as proof should receive the caveat: "Correlation is not causation."

> Second, SIDS prevalence data were collected from 15 countries, which reduced the power of our analyses and may have generated Type I/II errors. Third, pain management techniques practiced in various countries could not be accounted for in our study.

This is a significant admission. If pain causes trauma that leads to SIDS, then it stands to reason that effective pain management techniques should reduce or eliminate such a cause.

> Fourth, homogeneity of environmental exposure and diagnosis among the SIDS studies has been assumed, but each may be subject to misclassification. Fifth, we assumed the absence of neonatal female circumcision, which is illegal or uncommon in the studied countries. Six, the CDC lists SIDS for all autopsied and nonautopsied cases without distinction. In the case of an interracial parentage, the CDC only reports a single race, usually the one chosen by the mother.

Elhaid reported that the SIDS rate is 39% lower among Hispanics than non-Hispanics. And yet the percentage of multiracial infants in the United States in 2015 was 14% and growing. Interracial unions now account for 17% of all newlyweds and 10% of all married couples in the United States. And 42% of intermarried couples include a white partner and a Hispanic partner. So what effect does an increasing likelihood of multiracial parentaage have on the rates of SIDS by race? Elhaid doesn't explain.

> Some of the above-mentioned limitations were addressed by restricting our analyses to countries that perform autopsies and assembling a secondary dataset of US states. Although the age of inclusion for SIDS differs across countries, the difference centers on the inclusion of the first week of life, a time when a meager percentage of SIDS deaths occur [9, 34]. SIDS prevalence and the stressors' rates do not change dramatically over time [e.g., 9, 34, 105], thus accepting mismatched dates up a few years would likely have small effect on the results. A major difficulty is to find year-matched MNC and SIDS rates globally. We addressed this problem by deriving the low MNC rates from the proportion of Jewish and Muslims populations who tend to remain constant over short period of times and showed that halving or doubling their proportions does not change the results. Stang [106] found that most doctors and obstetricians who perform circumcisions avoid using anesthesia

Stang conducted a survey by mail in 1996 and reported that 45% of doctors used anesthesia. In the time since that survey was conducted, the AAP recommended in 1999 – and reiterated in 2012 – that all patients undergoing circumcision should be provided adequate analgesia. And Yawman found that by 2003, some 97% of residency programs were teaching the use of anesthesia for circumcisions. So it's likely that most doctors trained in the United States *do* use anesthesia. Elhaid's admission that he could not account for pain management techniques represents another serious limitation. Patients today are

likely receive some form of pain relief that would alleviate the types of stressful stimuli that Elhaik claims can cause SIDS.

> due to the extended time the procedure requires (half-hour)

Times vary. Several physicians have said that they perform a circumcision in just a few minutes.

> and its potentially negative effects [107-109].

Hermann [107] reported on the effects of "*repeated* pain experiences in neonates." Yet circumcision is normally a singular event. Fan [108] measured the sensitivity of Chinese men diagnosed with autism using images that depicted people in painful situations. Ing [109] studied whether circumcision performed *outside the perinatal period* was associated with an increased risk of a mental disorder diagnosis. It's unclear how these studies can shed light on negative effects of anesthesia to neonates.

> Some of the remaining limitations may be addressed in a carefully constructed cohort studies, but it is likely that other limitations cannot be addressed, in which case our confidence in the associations depends their replicability.

Conclusion

> SIDS is a diagnosis with a multifactorial underlying etiology. The allostatic load hypothesis [5] explains the main characteristics of SIDS (male predominance, different rates among US group, prevalence peak between 2 and 4 months, and seasonal variation) in the prolonged and repetitive stressful, painful, and traumatic stimuli that may begin prenatally, tax neonatal regulatory systems, and increase the risk of SIDS. Our ecological analyses support an association between MNC, prematurity, and SIDS and the additive effects of MNC and prematurity toward SIDS. Mitigating these and other stressors may reduce the prevalence of SIDS. Our data and code can be used to evaluate associations with other environmental factors. Future cohort studies should consider the existence of these stressors, genetic vulnerabilities, and life history.

Incredibly, the researcher did not identify a single case of circumcision causing a death from SIDS. His entire argument is based on a correlation of the rates of newborn circumcision with the rates of deaths attributed to SIDS.

In arriving at his conclusions, the researcher made several false or unsuppported assumptions. For example, he falsely assumed that the newborn circumcision rate among Muslims is 100%. He failed to account for the increasing use of pain relief. He cited studies of low quality, studies that are not relevant, and studies that don't support his claims.

"Hugo Heymans, one of the Netherlands' foremost pediatricians who for decades had worked at the Amsterdam Academic Medical Center, dismissed Elhaik's study as "flawed, biased and unreliable" … Heymans noted that Elhaik's study does not take into account the potential impact of additional factors that may influence SIDS. 'There are many social-economic differences between Hispanics and White Americans, as well as different eating habits' that are not factored in Elhaik's study, Heymans said."

Consequently this study cannot provide any evidence that circumcision affects the risk of SIDS.