Features

The Microethics of Informed Consent for Early Feminizing Surgery in Congenital Adrenal Hyperplasia

Katharine B. Dalke and Arlene B. Baratz

Ethics is what happens in every interaction between every doctor and every patient.
—Paul A. Komesaroff

ABSTRACT

Early surgery for genital difference in 46,XX congenital adrenal hyperplasia (CAH) is highly controversial, with contested evidence of benefits and risks. While professional urological societies and a parent-led CAH advocacy group maintain that families should have the option to consent for surgery for their child, former patients, intersex-led advocacy groups, and human rights and medical organizations denounce surgery on unconsenting infants for non-life-threatening genital variations. In the absence of clear data, clinicians are encouraged to engage in shared decision making with parents to obtain their fully informed consent.

Unexplored microethics issues regarding clinicians' implicit bias for treatments may interfere with their ability to obtain parents' fully informed consent in this setting. Implicit bias may be inferred from parents' experiences and from official and unofficial communications from clinicians.

Katharine Dalke, MD, MBE, is an Assistant Professor in the Department of Psychiatry and Behavioral Health at the Pennsylvania State University, Penn State College of Medicine, in Harrisburg, Pennsylvania. *kdalke@pennstatehealth.psu.edu*

Arlene B. Baratz, MD, is Coordinator of Medical and Research Affairs for InterConnect Support Group. *arlene@interconnect.support*

People with intersex traits/differences of sexual development (DSD) are born with sex traits, including chromosomes, gonads, or genitalia, that do not align with binary medical definitions of sex. Unless these differences are associated with urinary obstruction or exposed abdominal organs, these traits pose no immediate threat to physical health. One of the most common conditions associated with intersex traits is classical 46,XX congenital adrenal hyperplasia (CAH). Due to prenatal androgen accumulation, CAH may be associated with varying degrees of fusion of the urethra and vagina, clitoral enlargement, or development of a typical-appearing penis.[1] These variations are not dangerous, although associated adrenal enzyme deficiency can cause potentially life-threatening illness by blocking the production of hormones that regulate the immune system and fluid balance.

Early feminizing genital surgery seeks to normalize the appearance of the genitals in children who are assigned female gender by reducing clitoral size and creating an external vaginal opening. These surgeries have been performed on hundreds of infants under the age of two;[2] a review of two national billing databases, from 2009 to 2012 and 2004 to 2014, indicates that, during those periods, 291 and 544 female-assigned 46,XX children with CAH, respectively, underwent genital surgery in infancy.[3] Importantly, the cited aims of early genital surgery often combine physical and psychological goals. These include reducing urinary infections and incontinence, creating genital anatomy capable of future penetrative intercourse, preserving erogenous sensitivity, facilitating future reproduction, avoiding fluid or blood retention in the vagina and uterus, reducing the risk of stigma, promoting gender identity development "by providing anatomy concordant with gender of living," and responding "to parents' wishes for their child."[4]

Questions regarding the efficacy of these surgeries persist. There is some evidence that early feminizing genital surgery may accomplish its physical goals and can be satisfactory to some patients, but there is no compelling evidence that early surgery is superior or not inferior to deferral and the implementation of psychosocial interventions.[5]

There are also widely known and well-documented risks and complications of feminizing genital surgery in infancy. These include the harms of anesthesia, vascular injury, bleeding, infection, nerve damage, frequent re-operation, clitoral re-enlargement, vaginal stenosis, urinary pain/infection/incontinence, sexual dysfunction/impaired sensation, and permanent discordance of genital appearance with self-assigned gender.[6] Researchers have also observed that, in its confirmation that a person's natural genitals are abnormal, surgery may unintentionally compound shame and stigma.[7] Intersex advocates have brought personal experiences of these often irreversible consequences to the attention of medical and human rights organizations that support a child's right to autonomy, integrity, and health, several of which have concluded that the evidence of benefit is insufficient to outweigh the risks of genital surgery when a child is too young to give consent.[8]

In the face of limited and conflicting evidence regarding early feminizing genital surgery, as clinicians have observed, the usual clinical approach to treatment using "evidence-based medicine meets major hurdles."[9] Genital surgery is a topic of ongoing intense ethical debate.[10] Increasingly polar positions have been taken, ranging from deferral to parents' "absolute final right"[11] to make surgical decisions for their child, to legislative efforts to delay surgery until a child is old enough to give informed consent.[12]

In response to this debate, the Council on Ethical and Judicial Affairs of the American Medical Association recognized that parents are responsible for nurturing their child's health, well-being, autonomy, and personhood and concluded, "When no single approach can be said *a priori* to be 'best,' ethically sound practice requires that decisions be carefully tailored for each patient in a process of shared decision making among parents/guardians, physician and the patient (in keeping with the child's capacity to participate)."[13] This framing presents the decision for or against early feminizing genital surgery as a matter of equipoise, in which the risks and benefits of both options are equally medically sound and ethically viable, with shared decision making by the physician and parents the apparently ideal means to ensure informed consent.

The presumption of equipoise and the assumption that shared decision making is the path to an ethical and fully informed treatment plan are reflected in the statements of surgeons and surgical societies. The Societies for Pediatric Urology (SPU) states it is "not pro-surgery any more than . . . anti-surgery,"[14] and "supports parental and patient education with accurate evidence-based literature regarding the risks and benefits of all management options."[15] Statements by the SPU and the American Urological Association both speak to the necessity of fully informed consent, including freedom from coercion, the capacity for decision making, and the disclosure of all relevant information regarding the risks and benefits of all options, prior to surgery.[16]

The aim of this discussion is not to question whether clinical teams obtain informed consent in the macro ethical sense. Rather, we present evidence from the medical literature and practitioners' narratives that allows us to explore the microethics of obtaining informed consent for early feminizing genital surgery. Truog and colleagues described microethics as "the ethics of everyday practice."[17] In the present context, microethics would address these concerns: How do clinicians fulfill their obligation to inform parents within a given doctor-patient relationship? What information is presented, and in what manner? We further suggest that practitioners' implicit bias, from which no clinician is free and which tends to lead clinicians to recommend procedures they were trained to deliver,[18] is a driver in the microethics choices they make during informed consent discussions. Particularly in the pediatric context, these questions are crucial: informed consent, essential to the idea that parents have the right to make decisions in the best interest of their child, may be the victim of its own success, if clinicians fail to recognize the communication factors that compromise parents' ability to provide it.[19] This may become dire in the case of early feminizing genital surgery, in which "opinions and emotions may oust facts and foster confusion. . . . increased by the limits and pitfalls of 'evidence-based medicine,' a methodoloy that may be incompatible with some ethical considerations, leaving 'experience-based medicine' as the only tool."[20]

Given that this article centers on discussions of implicit bias, we would like to make explicit our own. As an intersex person and a parent of intersex children, and as active participants in intersex advocacy and support for a combined four decades, we are biased by our experiences and those of the intersex adults and families we've met to believe that intersex traits are healthy variations of human development. We are also biased to conclude that affirming psychosocial support is just as, and likely more, effective than surgical intervention in infancy to promote social, emotional, and sexual health and thriving. As physicians, we also hold the bias that physicians seek to do what is best for their patients, especially when what is best is scientifically ambiguous. We aim to balance these first biases with the last, and approach the following discussion with intellectual humility.

It is in the compromises struck by clinicians in the education space that we may infer their biases about what is important for parents to know, and how to interpret these compromises.

There is evidence that surgeons of all disciplines are biased toward surgery, even when the evidence base supports less-invasive treatment.[21] Regarding early feminizing genital surgery for CAH, one recent study in which more than half of the children in the study had CAH found a discrepancy in preoperative satisfaction with genital appearance: 30 percent of mothers and 50 percent of fathers were satisfied with their child's genital appearance, while 0 percent of the surgeons in the study were.[22] The authors observed that this discrepancy in pre-operative satisfaction may have "represented [surgeons'] unconscious bias to justify the surgery that they anticipated performing." Remarkably, 96 percent of the families in the study gave consent for genital surgery. Rates of consent that parallel surgeons' rather than parents' attitudes suggest that clinicians' attitudes and biases regarding a child's genitals may influence parents' decisions.

Current clinical practice and clinicians' communications suggest that clinicians' implicit bias may influence what and how parents learn about treatment alternatives. The goal of the process of parental education is help parents to achieve an understanding of the choices, risks, benefits, and alternatives they have, and to help them make decisions that are concordant with their family's values and preferences.[23] Ideally, this education includes a thorough and thoughtful review of the information described above. However, parents' informed consent may be compromised by the receipt of too much information.[24] When individuals receive large quantities of information, especially when it is ambiguous or contradictory, they may react negatively, experience higher levels of confusion, and narrow their choices more hastily,[25] which may cause emotional distress that may influence their decisions.[26]

Parents of a child with genital difference are at risk of isolation, confusion, and distress at levels comparable to parents of a child with a chronic illness.[27] This distress may further complicate a family's ability to deal with vast quantities of complex information, create cognitive overload, impair cognition, prevent full comprehension of the risks surrounding early feminizing genital surgery, and thereby overwhelm their capacity to make decisions.[28] The option to exercise therapeutic privilege, that is, to totally withhold distressing diagnostic information from a family in the interest of preserving their autonomy, has happily not been standard practice since a 2005 Consensus Statement by the International Consensus Conference on Intersex recommended routine disclosure.[29]

However, clinicians' assessment of how to communicate the right information to a family in the right way, to facilitate their decision making,[30] is a process of micro ethics. It is in the compromises struck by clinicians in the education space that we may infer their biases about what is important for parents to know, and how to interpret these compromises.

Investigation of clinical informed consent protocols, for example, indicates that parental education is often incomplete. In a study of DSD clinics, fewer than one-third of the staffs at the clinics reported they documented discussions of risks with parents that included the need for additional procedures and effects of the interventions on sexual function, and one-sixth or fewer of the staffs reported that they discussed potential psychological effects, gender uncertainty, reversibility, and the elective nature of interventions with parents.[31]

In another study concerning medical decision making, 41 percent of parents said that they did not understand their child's DSD diagnosis.[32] In interviews, parents who chose surgery said that their doctors had given them an incomplete picture of the risks and alternatives, or had even provided misleading information.[33] The consequences of providing inadequate information are significant: not only can the process of obtaining fully informed consent be undermined in the immediate term, but incomplete counseling will leave parents poorly prepared to deal with the implications of their decisions in the future.

Because it appears that few care teams discuss the elective nature of early feminizing genital surgery, parents who consider infant surgery may not understand that procedures like clitoroplasty (surgery to reshape the clitoris and make it smaller) and labiaplasty (surgery to reshape the labia to appear more "feminine") are not medically necessary.[34] Interventions to change a physical state such as genital variation may be considered necessary when that state poses a serious, time-sensitive threat to health, such as functional impairment, and the intervention is the least harmful way to alleviate that threat.[35] When genital difference itself poses no immediate threat to a child's physical well-being, intervention for that difference is not medically necessary. The SPU, however, argues that surgeons include "emotional concerns" in discussions of medical necessity.[36] Despite a specific 2015 World Health Organization (WHO) designation of genital surgery as "medically unnecessary,"[37] some contend that "medically unnecessary is too narrow to use in this complex-patient population because the WHO [in 1946][38] defines health in encompassing psychological, psychosocial and developmental health."[39]

This psychosocial argument for physical intervention reveals a core implicit bias, that "body differences associated with DSDs may harm well-being."[40] This bias has been stated explicitly by the European Society of Pediatric Urology, which asserted, "Atypically developed genitalia can affect not only physical appearance and body image, but also . . . psychological and psychosexual development of the individual."[41] A past president of the SPU stated, "We're not saying that there is definitely going to be a negative outcome psychologically of having ambiguity, but [there are] several conditions or outcomes that can happen if surgery doesn't happen, if nothing is done, and people have the normal sexual urges that they may have. Then it's as any sexual interaction in someone who has not had surgery, the negative impact is significant."[42] In addition to presumed stigma, some have cited high rates of suicide attempts among transgender youth as evidence that living with genital variation confers a 40 percent risk of suicide.[43] Some have suggested that genital variation has wide-ranging effects that could even preclude a person from becoming a functioning member of society: "There is far more psychological impairment if you don't let genetic females be raised to their fullest potential."[44] With surgery, another surgeon avers, they "can have great jobs, ultimately pay taxes, and be part of a community."[45]

These statements are inconsistent with the available evidence. A 2016 update of the 2005 Consensus Statement by the International Consensus Conference on Intersex reports "there is no evidence regarding the impact of surgically treated or non-treated DSDs during childhood for the individual, the parents, society or the risk of stigmatization."[46] An association between genital difference and psychosocial distress has never been convincingly demonstrated.[47] Recent evidence regarding stigma specifically in adult women with CAH suggests that when they do experience social stigma, it is in relation to behavioral or external physical difference rather than their genital variation;[48] stigma in sexual settings occurs regardless of whether they have had surgery or not.[49] While there have been few studies regarding the number of families who consent to surgery for their child, in 2016 Nokoff and colleagues reported that 96 percent of families in their study consented to surgery.[50] If this is indicative of the rate of consent to surgery, then, as the update to the 2005 Consensus Statement noted, "the high prevalence of normalizing surgery makes it impossible to separate the differences and surgical management."[51] Regardless, significant bias remains, and failure to conform to gender and sexual norms and expectations can compromise emotional health to such an extent that a pediatric urologist characterized decisions about early genital surgery as "the most consequential health decisions of [families'] lives."[52]

The bias that psychological discomfort is inevitable and medically treatable can tip the balance toward intervention without decision makers being aware this is happening,[53] an effect that has been underestimated by clinicians.[54] Indeed, many families feel pressured to accept irreversible procedures, and may believe that surgery will offer swift relief for their anxiety.[55] Reduction of parents' distress is often cited and studied as an outcome of early genital surgery, with some evidence that parental distress does decrease after surgery.[56] It remains unknown whether symptom resolution is related to surgery or is a function of adaptation, such that parents whose children do not undergo surgery would also experience similar improvement.[57] Nevertheless, the appeal of a quick fix is strong, especially for distressed parents. Having expectations that surgery can fix anything,[58] stressed parents who fear for their child may be easily convinced that surgery should not be deferred.

Although psychosocial interventions are recommended as a primary means to treat distress, and a recent consensus group concluded that psychosocial care for children and families is obligatory,[59] a lack of validated interventions and specialized care-providers limits availability.[60] Furthermore, the assumption that surgery can summarily avert psychosocial distress may in turn undervalue psychological support. As a mental health practitioner said, "Part of my job on the DSD team is to convince the other team members that there's a crucial role for mental health. In my first year on the team I was accidentally forwarded an email thread in which a few of the surgeons questioned why we even needed a mental health specialist."[61] Peer support, which can help reduce parents' fears and promote coping,[62] is often underutilized.[63] Even the professional surgical standards indicate that support is beneficial primarily as a pathway to, rather than around, surgery, advocating "complete informed consent with counseling and support . . . prior to proceeding with any surgical intervention."[64]

The assumption that surgery is a more effective intervention than psychosocial support lacks robust evidence.[65] To the contrary, there is evidence that deferral of surgery with psychological support for families and children has positive outcomes. Parents who rejected early genital surgery for various DSD said their child had not experienced unusual bullying or harassment related to the child's difference; their child went to school and had friends like other children.[66] In a feasibility study of seven French families of children with CAH who opted for ongoing psychological support and endocrine treatment, genital difference decreased significantly with time, and "girls and their parents have not experienced significant concerns regarding genital ambiguity."[67] The outcome of this implicit bias is that families may be led to believe that their options are to consent to early feminizing genital surgery or to do nothing, leaving them "between a rock and a hard place."[68]

In addition to the necessity and benefit of surgery, there is also evidence that recommendations regarding the timing of surgery may be influenced by implicit bias. Emblematic of this are claims that "nearly 90 percent of CAH patients believe that the procedure should be done within the first year of life"[69] and that "the vast majority are overwhelmingly happy that they had it as a baby."[70] Perhaps not coincidentally, urologic surgeons have a strong bias towards performing surgery early: in one survey of pediatric urologists, 78 percent of respondents preferred early surgery, citing factors such as better quality of genital tissue in infants and expectations that only minor procedures would be required later.[71] As an individual surgeon remarked, "there's clear data that doing these surgeries younger are better"[72] for outcomes and patients' satisfaction. When confronted with long-term data regarding poor outcomes from early surgery, surgeons are confident that "surgical techniques continue to improve and the scientific literature reports current outcomes to be significantly better than in prior generations,"[73] and that "we know that we're so much better now than we were 30, 40 years ago . . . it's much harder for us to accept that argument that things are bad."[74]

The oft-repeated contention that patients prefer earlier timing relies on the conclusions of very few studies, and is contradicted by close scrutiny of the study methods and data.[75] In these studies, including a 2016 French study,[76] those who "preferred" early surgery were not informed of the alternative of not having surgery at all; that delaying surgery could have reduced the rate of re-operation for vaginal stenosis; or that there had been significant technical modernizations predicted to improve outcomes since their early childhood surgery.[77]

Additionally, numerous authors acknowledge the lack of evidentiary support for the superiority of early versus deferred surgery.[78] As Creighton writes, "the ideal timing and nature of surgical reconstruction . . . is highly controversial . . . evidence-based recommendations still cannot be made."[79] In the absence of research that directly compares the outcomes of early and later feminizing genital surgery, it is impossible to say which is superior. Some gynecologists who perform both early and postpubertal surgery advocate for deferral because they observe better healing in the presence of endogenous estrogen,[80] and a recent case report described a good outcome and preservation of sexual function following feminizing genital surgery in an adult woman with CAH, which was performed by a surgeon with expertise in surgery for adults.[81] Although contemporary surgical procedures may yield better outcomes than the clitoral amputations of the distant past, "there is controversy on functional outcome of clitoral surgery despite using modern techniques . . . [which do] not necessarily assure well sexual function. . . . While most surgeons caring for these patients claim they achieve 'good cosmetic results' the long-term functional outcomes are scant and mostly disappointing."[82] Despite this evidence, pediatric urologists may retain their bias towards early surgery in part because of a lack of training to perform surgery in older, consenting individuals,[83] and informed consent may be further compromised by the irrelevance of long-term medical consequences of modern early feminizing genital surgery, the data for which will have been made obsolete by newer surgical techniques by the time information is available in 15 or 20 years.[84]

Clinicians tend to cite data drawn from older studies that relied on now-invalid methods, or that conflated gender behavior with identity.

Satisfaction with assigned gender in CAH is an important outcome for families who consider surgery to align genital anatomy with gender assignment. Because the infants have XX chromosomes, a uterus, and ovaries, female gender assignment is generally recommended.[85] Interestingly, evidence suggests that clinicians may underreport the risk of performing gender-assigning surgery on someone who grows up to not identify as female. Clinicians tend to cite data drawn from older studies that relied on now-invalid methods, or that conflated gender behavior with identity.[86] Based on these studies, it is commonly cited in the literature that the rate at which recipients of early surgery do not later identify as female is 5 to 10 percent,[87] and these data are quoted to parents regarding the risks of performing early surgery. Further, some surgeons have articulated a bias that could be described as cisnormative: that is, even though "gender identity [is] defined by the individual,"[88] "if you have two X chromosomes, you're genetically female."[89] (*Cis* refers to a sense of gender identity that corresponds with one's birth sex. In the setting of a child with CAH where genitalia are not typically male or female and chromosomes are XX, female gender identity is presumed.) Despite finding that 14 percent of CAH adults identify as intersex rather than female, a 2016 study stated, "for physicians it is obvious and unequivocal that a person with [46,XX CAH] has a female gender identity."[90] Whatever the genital appearance, according to one surgeon, "these patients with Congenital Adrenal Hyperplasia are female, and they are not born with a penis, they are born with an enlarged clitoris, and sometimes the clitoris looks like a penis, and it could be four to five inches long."[91]

These assumptions are contradicted by the available data. The only study that used psychiatric diagnostic criteria reported that 13 percent of the four- to 11-year-old subjects of the study exhibited cross-gender behavior that met those criteria sufficiently to stimulate referral to a gender clinic.[92] A recent systematic review and meta-analysis reports that, in two studies, 63 of 71 subjects (88.7 percent) of 46,XX subjects with CAH who had feminizing genital surgery identified as female, and therefore 11.3 percent did not identify as female; data from additional three studies indicate that 15 of 115 subjects (13 percent) identified as "mixed" gender.[93] These data must be considered on the background of the prevalence of transgender identity in the general population, which has been estimated to be 0.6 percent based on data from the Behavioral Risk Factor Surveillance Survey.[94]

The consequences of minimizing these risks can be catastrophic: incongruence between gender identity and gender assignment can cause gender dysphoria, which has been associated, in transgender individuals, with severe psychological distress and disproportionate rates of psychiatric illness and suicidality.[95] The distress of gender dysphoria in individuals with DSD can be relieved by affirming a person’s gender, sometimes including social and physical gender transition, which has been reported to dramatically reduce elevated rates of psychiatric illness and psychological distress.[96] In CAH, because early feminizing genital surgery is performed to reinforce female assignment long before a child expresses gender identity, genital structures may be irreversibly removed that align with eventual adult gender identity, that could limit a person’s later options for gender affirmation. Because there is no reported way to predict gender identity, whether an adult will be satisfied with early surgery that was performed to provide “correct” genitalia is a gamble. As one endocrinologist said, “When we’re trying to force people into cultural normative, hetero-normative situations, there’s a high chance that we’re going to make some major mistakes and harm people irreparably.”[97] And yet, decisions regarding surgery appear to occur in the setting of strong bias that a child with CAH is destined to be female, and that surgery can not only “normalize” genitals, but serve as a sex-gender realignment technique.[98] Furthermore, interviews suggest that families are introduced to the bias that surgery is *required* as part of effective sex assignment.[99]

In CAH, because early feminizing genital surgery is performed to reinforce female assignment long before a child expresses gender identity, genital structures may be irreversibly removed that align with eventual adult gender identity, that could limit a person’s later options for gender affirmation.

A multidisciplinary team model is thought to optimize the quality of care, with “the evaluation and management of each child individualized and undertaken using a multi-specialty and family-centered approach,”[100] and is frequently held up as the standard of care. However, decision making within a multidisciplinary team structure remains susceptible to bias. An analysis of interactions among DSD team members and families found that clinicians strategically deployed uncertainty to steer parents to the perceived clinically appropriate option.[101] Even when surgeons urge caution, emphasize that surgery is elective and controversial, and discuss the option of not doing surgery, to also tell parents that genital difference can be stigmatizing and to present surgery as a relatively straightforward way to make genitalia more gender-typical implies that surgery can deter stigma and so should be done sooner rather than later.[102] While ostensibly offering information and choice, the selected information that is presented to parents reveals biases that genital difference is abnormal, dangerous, and more easily and effectively treated by surgery than psychosocial support, especially when done early. The result is a mixed message that covertly conveys the medical necessity of surgery.[103] Evidence exists that physicians sometimes deliberately manipulate their power to yield the decision that they believe is correct.[104] One endocrinologist on a team said that while she had advised 240 families over three years to delay surgery, not one had taken her advice: “This decision is made in the end with our surgeons. . . . If a family wants a surgery, [the surgeons are] much less likely than non-surgeons to say, ‘no we’re not going to do that.’ ”[105] Within the multidisciplinary team, the microethics of what to discuss with families and how to discuss it can intersect with the team’s power structure to bias decision making towards a surgical intervention.

An example of a balanced family education approach that avoids these issues of microethics is detailed in a recently published shared decision-making checklist for early feminizing genital surgery in CAH, at Phoenix Children’s Hospital.[106] It includes suggestions from patient advocates and discussion of various topics during multiple visits, in which parents’ understanding is confirmed using a teach-back method. Part 1 of the checklist is an overview of the diagnosis and treatment goals. Part 2 addresses preferred language regarding anatomy and diagnosis. Part 3 explores parents’ preferences for information and addresses topics for review over multiple visits including changes in puberty, sexual health, gender identity, fertility, and genetics. Part 4 suggests questions about ongoing care regarding medication and treatment and provides information about support and advocacy groups. Part 5 explores issues of body image and addresses the importance of professional mental health support. Part 6 focuses on surgery: the first portion addresses surgical procedures, irreversibility, controversies, and risks and benefits; next, deferring or forgoing surgery are discussed, and families are required to talk to people with CAH who have chosen and declined surgery to learn about their real-life experiences. In early experience, three of four families who used the checklist deferred surgery.[107]

In conclusion, even in the presence of the macroethics elements of obtaining fully informed consent, implicit bias may influence the microethics of shared decision making and compromise parents' informed consent for early feminizing genital surgery in CAH. These include biases that genital difference is incompatible with the well-being of the child and poses a threat to mental health; that surgery effectively averts psychosocial distress and the need for psychosocial support; that early surgery is better and more effective than later surgery; that people with CAH are always female; that gender dysphoria is rare; and that surgery is necessary to assign gender.

Furthermore, there is emerging evidence that these biases and team power dynamics can erode the ability of multidisciplinary teams to provide collaborative care and ensure shared decision making with parents. A newly created checklist for decision making over multiple visits that has enabled parents to defer surgery includes comprehensive information, the lived experience of adults who did and did not have surgery, and peer and psychosocial support. Further research is needed to directly assess the presence and impact of biases held by parents and clinicians, and to explore the role of interventions that promote more effective shared decision making.

NOTES

The quotation from Paul A. Komesaroff at the beginning of this article is from P.A. Komesaroff, *Troubled Bodies: Critical Perspectives on Postmodernism, Medical Ethics, and the Body* (Durham, N.C.: Duke University Press, 1995).

1. P.A. Komesaroff, *Troubled Bodies: Critical Perspectives on Postmodernism, Medical Ethics, and the Body* (Durham, N.C.: Duke University Press, 1995).

2. A.P. Hardin, J.M. Hackell, and the AAP Committee on Practice and Ambulatory Medicine, "Age Limit of Pediatrics," *Pediatrics* 140, no. 3 (13 April 2017), *https://pediatrics.aappublications.org/content/pediatrics/140/3/e20172151.full.pdf.*

3. R.M. Sturm, B. Durbin-Johnson, and E.A. Kurzrock, "Congenital adrenal hyperplasia: Current surgical management at academic medical centers in the United States," *Journal of Urology* 193, no. 5 (2015): 1796-801; J.D. Roth et al., "Characteristics of Female Genital Restoration Surgery for Congenital Adrenal Hyperplastia Using a Large-Scale Administrative Database," *Urology* 115 (2018): 162-7.

4. D.B. Gorduza, C.A. Quigley, A.A. Caldamone, and P.D.E. Mouriquand, "Surgery of Anomalies of Gonadal and Genital Development in the 'Post-Truth Era,' " in *Pediatric Urology, an Issue of Urologic Clinics,* ed. H.L. Copp, A.J. Lorenzo, A.R. Shukla, and A. Caldamone (New York: Elsevier, 2018),659-69.

5. M.J. Elders, D. Satcher, and R. Carmona, "Re-Thinking Genital Surgeries on Intersex Infants," *Palm Center Blueprints for Sound Public Policy,* 2017, *https://www.palmcenter.org/wp-content/uploads/2017/06/Re-Thinking-Genital-Surgeries-1.pdf.*

6. M. Kaefer and R.C. Rink, "Treatment of the Enlarged Clitoris," *Frontiers in Pediatrics* 5 (2017): 125, *https://www.ncbi.nlm.nih.gov/pmc/articles/PMC5581357/*; L.C. Wang and D.P. Poppas, "Surgical outcomes and complications of reconstructive surgery in the female congenital adrenal hyperplasia patient: What every endocrinologist should know," *Journal of Steroid Biochemistry and Molecular Biology* 165, pt. A (2017): 137-44; P.P. Dangle et al., "Surgical Complications Following Early Genitourinary Reconstructive Surgery for Congenital Adrenal Hyperplasia-Interim Analysis at 6 Years," *Urology* 101 (2017): 111-5; J. Stites et al., "Urinary continence outcomes following vaginoplasty in patients with congenital adrenal hyperplasia," *Journal of Pediatric Urology* 13, no. 1 (2017): 38 e1-7; M.C. Davies, N.S. Crouch, C.R.J. Woodhouse, and S.M. Creighton, "Congenital adrenal hyperplasia and lower urinary tract symptoms," *BJU International* 95, no. 9 (2005): 1263-6.

7. K. Roen and P. Hegarty, "Shaping parents, shaping penises: How medical teams frame parents' decisions in response to hypospadias," *British Journal of Health Psychology* 23, no. 4 (November 2018): 967-81.

8. Elders, Satcher, and Carmona, "Re-Thinking Genital Surgeries," see note 5 above; U.S. Bureau of Public Affairs, "In Recognition of Intersex Awareness Day," 25 October 2017, *https://2009-2017.state.gov/r/pa/prs/ps/2016/10/263578.htm;* German Ethics Council, "Intersex people should be recognized, supported and protected from discrimination," 2012, *https://www.ethikrat.org/en/press-releases/2012/german-ethics-council-intersex-people-should-be-recognized-supported-and-protected-from-discrimination/?cookieLevel=not-set&cHash=f60c9211b61ad2ff788bf7482969b200;* Swiss National Advisory Committee on Biomedical Ethics, "On the management of Difference of Sex Development: Ethical Issues Relating to Intersexuality: Opinion number 20/2012," 2012, *https://www.ilga-/europe.org/sites/default/files/swiss_ethics_council_ccl_2012.pdf;* Amnesty Interna-

tional, "First do no harm: ensuring the rights of children born intersex," 9 May 2017, *https://www.amnesty.org/en/latest/campaigns/2017/05/intersex-rights/;* Human Rights Watch, " 'I Want to be Like Nature Made Me': Medically Unnecessary Surgeries on Intersex Children in the US," video, 2017, *https://www.hrw.org/report/2017/07/25/i-want-be-nature-made-me/medically-unnecessary-surgeries-intersex-children-us#;* Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," 20 October 2017, *https://phr.org/news/unnecessary-surgery-on-intersex-children-must-stop/;* J. Toler, "Medical and Surgical Intervention of Patients with Differences in Sex Development," GLMA Policy and Government Affairs Committee, 2016, *http://glma.org/index.cfm?fuseaction=document.viewdocument&ID=CEB9FEE4B8DD8B7F4F7575376BD476C3A433379DD853BEA1 7913AFCCB8270299C0731320B03D2F5E1022F1C15602FBEA;* American Medical Student Association, "AMSA Issues Statement to Defer Gender 'Normalizing' Surgeries for Children Born as Intersex," 26 October 2017, *https://www.intersexequality.com/amsa-issues-statement-to-defer-gender-normalizing-surgeries-on-intersex-children/;* American Academy of Family Physicians, "Genital Surgeries in Intersex Children," July 2018, *https://www.aafp.org/about/policies/all/genital-surgeries.html.*

9. P.D Mouriquand et al., "Surgery in disorders of sex development (DSD) with a gender issue: If (why), when, and how?" *Journal of Pediatric Urology* 12, no. 3 (2016): 139-49.

10. J.A. Greenberg, "Legal, ethical, and human rights considerations for physicians treating children with atypical or ambiguous genitalia," *Seminars in Perinatology* 41, no. 4 (June 2017): 252-5; L.H. Gillam, J.K. Hewitt, and G.L. Warne, "Ethical principles for the management of infants with disorders of sex development," *Hormones Research in Paediatrics* 74, no. 6 (2010): 412-8; M. Carpenter, "The 'Normalization' of Intersex Bodies and 'Othering' of Intersex Identities in Australia," *Journal of Bioethical Inquiry* 15, no. 4 (December 2018): 487-95; A.A. Kon, "Ethical issues in decision-making for infants with disorders of sex development," *Hormone and Metabolic Research* 47, no. 5 (2015): 340-3.

11. H. Scherz, "Parental rights," The Doctor's Lounge, America's Web Radio, 10 January 2019, podcast, *https://www.americaswebradio.com/podcasts/DoctorsLoungeJan10.2019.mp3.*

12. D. Stroumsa, "The state of transgender health care: Policy, law, and medical frameworks," *American Journal of Public Health* 104, no. 3 (2014): e31-8.

13. American Medical Association Council on Ethical and Judicial Affairs, "CEJA Report 3-I-18, Amendment to E-2.2.1, Pediatric Decision Making," 2018, *https://www.ama-assn.org/system/files/2019-12/i18-ceja-report-3.pdf.*

14. "Pediatric Decision Making and Differences of Sex Development: A Societies for Pediatric Urology and American Urological Association Joint Position Statement," July 2019, *https://www.auanet.org/guidelines/joint-statement-on-dsd.*

15. Ibid.

16. American Urological Association, "Code of Ethics," 2019, *https://www.auanet.org/myaua/aua-ethics/code-of-ethics;* Societies for Pediatric Urology, "Physicians recommend individualized, multi-disciplinary care for children born 'intersex,' " 11 November 2017, *http://www.spuonline.org/HRW-interACT-physicians-review/2017.*

17. R.D. Truog et al., "Microethics: The Ethics of Everyday Clinical Practice," *Hastings Center Report* 45, no. 1 (2015): 11-7.

18. S. Sah, A. Fagerlin, and P. Ubel, "Effect of physician disclosure of specialty bias on patient trust and treatment choice," *Proceedings of the National Academy of Sciences of the USA* 113, no. 27 (2016): 7465-9.

19. D. Hall, A. Prochazka, and A. Fink, "Informed consent for clinical treatment," *Canadian Medical Association Journal* 84, no. 5 (2012): 533-40.

20. Gorduza et al., "Surgery of Anomalies of Gonadal and Genital Development," see note 4 above.

21. Sah, Fagerlin, and Ubel, "Effect of physician disclosure of specialty bias," see note 18 above; Hall, Prochazka, and Fink, "Informed consent," see note 19 above.

22. N.J. Nokoff et al., "Prospective assessment of cosmesis before and after genital surgery," *Journal of Pediatric Urology* 13, no. 1 (2017): e1-28, e6.

23. Mouriquand et al., "Surgery in disorders of sex development," see note 9 above.

24. Human Rights Watch, "I Want to be Like Nature Made Me," see note 8 above.

25. C. Gebele et al., "Applying the concept of consumer confusion to healthcare: Development and validation of a patient confusion model," *Health Services Management Research* 27 no. 1-2 (2014): 1021.

26. E.A. Lipstein, W.B. Brinkman, and M.T. Britto, "What is known about parents' treatment decisions? A narrative review of pediatric decision making," *Medical Decision Making* 32, no. 2 (2012): 246-58.

27. A.B. Wisniewski, "Psychosocial implications of disorders of sex development treatment for parents," *Current Opinions in Urology* 27, no. 1 (2017): 11-3.

28. Gebele et al., "Applying the concept of consumer confusion," see note 25 above; J. Bester, C.M. Cole, and E. Kodish, "The Limits of Informed Consent for an Overwhelmed Patient: Clinicians' Role in Protecting Patients and Preventing Overwhelm,"*AMA Journal of Ethics* 18, no. 9 (2016): 869-86; A. Tamar-Mattis et al., "Emotionally and cognitively informed consent for clinical care for differences of sex development," *Psychology & Sexuality* 5, no. 1 (2013): 44-55; V. Pasterski et al., "Predictors of posttraumatic stress in parents of children diagnosed with a disorder of sex development," *Archives of Sexual Behavior* 43, no. 2 (2014): 369-75.

29. P.A. Lee et al., "Consensus statement on management of intersex disorders. International Consensus Conference on Intersex," *Pediatrics* 118, no. 2 (2006): e488-500.

30. D.L. Scammon et al., "Transforming consumer health," *Journal of Public Policy Market* 30, no. 1 (2011): 14-22.

31. A.M Rolston et al., "Disorders of sex development (DSD): Clinical service delivery in the United States," *American Journal of Medical Genetics Part C: Seminars in Medical Genetics* 175, no. 2 (2017): 268-78.

32. E. Bennecke et al., "Subjective need for psychological support (PsySupp) in parents of children and adolescents with disorders of sex development (dsd)," *European Journal of Pediatrics* 174, no. 10 (2015): 1287-97.

33. Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," see note 8 above.

34. K. Karkazis, A. Tamar-Mattis, and A.A. Kon, "Genital Surgery for Disorders of Sex Development: Implementing a Shared Decision-Making Approach," *Journal of Pediatric Endocrinology Metabolism* 23 (2010): 789-806.

35. B.D. Earp, T*he Child's Right to Bodily Integrity, in Ethics and the Contemporary World* (New York: Routledge, 2019).

36. Societies for Pediatric Urology, "Legislation Restricting Medical Options," 24 April 2019, *https://spuonline.org/Legislation.cgi.*

37. World Health Organization, "Sexual health, human rights and the law," 2015, *https://apps.who.int/iris/handle/10665/175556; W*orld Health Organization, "Sexual health, human rights and the law," 20 Feb 2020 *http://apps.who.int/iris/bitstream/handle/10665/175556/9789241564984_eng.pdf;jsessionid=0903E8FBEC246D7B5D8B6B8EEB5986B7?sequence=1.*

38. "Preamble to the Constitution of WHO as adopted by the International Health Conference, 19 June-22 July 1946; signed on 22 July 1946 by the representatives of 61 States, Official Records of World Health Organization, no. 2, p. 100, entered into force 7 April 1948, *https://apps.who.int/iris/bitstream/handle/10665/85573/Official_record2_eng.pdf?sequence=1.*

39. Hearings before the Connecticut Public Health Committee, "An Act Concerning A Person's Intersex Status or Characteristics," transcript, Senate Bill 388, 25 February 2019.

40. P.A. Lee et al., "Global Disorders of Sex Development Update since 2006: Perceptions, Approach and Care," *Hormone Research in Paediatrics* 85, no. 3 (2016): 158-80.

41. P. Mouriquand et al., "The ESPU/SPU standpoint on the surgical management of Disorders of Sex Development (DSD)," *Journal of Pediatric Urology* 10, no. 1 (2014): 8-10.

42. Scherz, "Parental rights," see note 11 above.

43. K. Knight, "If a Doctor Tells a Parent that Surgery Could Prevent their Child's Suicide, It's Not True," *Huffington Post,* 10 September 2017, *https://www.huffpost.com/entry/if-a-doctor-tells-a-parent-that-surgery-could-prevent_b_59af2445e4b0c50640cd62da.*

44. "Should California ban nonconsensual 'medically unnecessary' surgery on intersex minors?" *AirTalk, National Public Radio*, 5 February 2019, *https://www.scpr.org/programs/airtalk/2019/02/05/64157/should-ca-ban-nonconsensual-medically-unnecessary/.*

45. Ibid.

46. Lee et al., "Global Disorders of Sex Development," see note 40 above.

47. Elders, Satcher, and Carmona, "Re-Thinking Genital Surgeries," see note 5 above.

48. H.F.L. Meyer-Bahlburg et al., "Syndrome-Related Stigma in the General Social Environment as Reported by Women with Classical Congenital Adrenal Hyperplasia," *Archives of Sexual Behavior* 46, no. 2 (2017): 341-51.

49. H.F.L. Meyer-Bahlburg et al., "Stigma Associated with Classical Congenital Adrenal Hyperplasia in Women's Sexual Lives," *Archives of Sexual Behavior* 47, no. 4 (May 2018): 943-51.

50. Nokoff et al., "Prospective assessment of cosmesis before and after genital surgery," see note 22 above.

51. Lee et al., "Global Disorders of Sex Development," see note 40 above.

52. L. Palmer, "Guest Opinion: Intersex bill would harm those it aims to protect," *Bay Area Reporter,* 14 March 2019, *https://issuu.com/bayareareporter/docs/march_14__2019.*

53. J.C. Streuli et al., "Shaping parents: Impact of contrasting professional counseling on parents' decision making for children with disorders of sex development," *Journal of Sexual Medicine* 10, no. 8 (2013): 1953-60.

54. Roen and Hegarty, "Shaping parents," see note 7 above.

55. Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," see note 8 above.

56. Lee et al., "Consensus statement on management of intersex disorders," see note 29 above.

57. R.E. Ellens et al., "Psychological adjustment in parents of children born with atypical genitalia one year after their child undergoes genitoplasty," *Journal of Urology* 19, no. 4 (2017): 914-20.

58. Roen and Hegarty, "Shaping parents," see note 7 above.

59. S. Krege et al., "Variations of sex development: The first German interdisciplinary consensus paper," *Journal of Pediatric Urology* 25, no. 2 (April 2019): 114-23.

60. Rolston et al., "Disorders of sex development (DSD)," see note 31 above; M.M. Ernst et al., "Disorders of Sex Development/Intersex: Gaps in Psychosocial Care for Children," *Pediatrics* 142, no. 2 (2018): e20174045.

61. Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," see note 8 above.

62. Ibid., A.B. Baratz, M.K. Sharp, and D.E. Sandberg, "Disorders of sex development peer support," *Understanding Differences and Disorders of Sex Development* 27 (2014): 99-112; C.P. Houk and P.A. Lee, "Update on disorders of sex development," *Current Opinions on Endocrinology, Diabetes, and Obesity* 19, no. 1 (2012): 28-32.

63. E. Magritte, "Working together in placing the long term interests of the child at the heart of the DSD evaluation," *Journal of Pediatric Urology* 8, no. 6 (2012): 571-5.

64. Societies for Pediatric Urology, "Physicians recommend individualized, multi-disciplinary care," see note 16 above.

65. Elders, Satcher, and Carmona, "Re-Thinking Genital Surgeries on Intersex Infants," see note 5 above; Roen and Hegarty, "Shaping parents," see note 7 above; Ernst et al., "Disorders of Sex Development/Intersex," see note 60 above.

66. Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," see note 8 above.

67. P. Bougnères et al., "Deferring surgical treatment of ambiguous genitalia into adolescence in girls with 21-hydroxylase deficiency: A feasibility study," I*nternational Journal of Pediatric Endocrinology* 3 (2017): *https://doi.org/10.1186/s13633-016-0040-8.*

68. L.M. Liao, D. Wood, and S.M. Creighton "Parental choice on normalising cosmetic genital surgery," *BMJ* 351 (28 September 2015): h5124.

69. Palmer, "Guest Opinion: Intersex bill," see note 52 above.

70. Stroumsa, "The state of transgender health care," see note 12 above.

71. F. Yankovic et al., "Current practice in feminizing surgery for congenital adrenal hyperplasia: A specialist survey," *Journal of Pediatric Urology* 9, no. 6, pt B (2013): 1103-7.

72. Stroumsa, "The state of transgender health care," see note 12 above.

73. Societies for Pediatric Urology and American Urological Association, "Pediatric Decision Making," see note 14 above.

74. Stroumsa, "The state of transgender health care," see note 12 above.

75. K.M. Szymanski et al., "Parental decisional regret and views about optimal timing of female genital restoration surgery in congenital adrenal hyperplasia," *Journal of Pediatric Urology* 14, no. 2 (April 2017): 156.e1-e7; H.F.L. Meyer-Bahlburg, *Psychoendocrinology of Congenital Adrenal Hyperplasia in Genetic Steroid Disorder* (Boston: Elsevier, 2014), 285-300; H.F.L. Meyer-Bahlburg, "Misrepresentation of evidence favoring early normalizing surgery for atypical sex anatomies: Response to Baratz and Feder," *Archives of Sexual Behavior* 44, no. 7 (2015): 1765-8.

76. A. Binet et al., "Should we question early feminizing genitoplasty for patients with congenital adrenal hyperplasia and XX karyotype?" *Journal of Pediatric Surgery* 51, no. 3 (2016): 465-8.

77. Meyer-Bahlburg, "Misrepresentation," see note 75 above.

78. L.A. Piaggio, "Congenital Adrenal Hyperplasia: Review from a Surgeon's Perspective in the Beginning of the Twenty-First Century," *Frontiers in Pediatrics* 1 (2014): 50; K.P. Wolffenbuttel and N.S. Crouch, "Timing of feminising surgery in disorders of sex development," *Endocrine Development* 27 (2014): 210-21; W.L. Lean et al., "Cosmetic and anatomic outcomes after feminizing surgery for ambiguous genitalia," *Journal of Pediatric Surgery* 40, no. 12 (2005): 1856-60; L.H. Braga and J.L. Pippi Salle, "Congenital adrenal hyperplasia: A critical appraisal of the evolution of feminizing genitoplasty and the controversies surrounding gender reassignment," *European Journal of Pediatric Surgery* 19, no. 4 (2009): 203-10; F. Eckoldt-Wolke, "Timing of surgery for feminizing genitoplasty in patients suffering from congenital adrenal hyperplasia," *Endocrine Development* 27 (2014): 203e9.

79. S. Creighton et al., "Timing and nature of reconstructive surgery for disorders of sex development: Introduction," *Journal of Pediatric Urology* 8, no. 6 (2012): 602-10.

80. Bougnères et al., "Deferring surgical treatment of ambiguous genitalia," see note 67 above; Creighton et al., "Timing and nature of reconstructive surgery," see note 79 above.

81. W.A.A. Tjalma, "Assembling a Functional Clitoris and Vulva from a Pseudo-Penis: A Surgical Technique for an Adult Woman with Congenital Adrenal Hyperplasia," *Journal of Pediatric Adolescent Gynecology* 30, no. 3 (2017): 425-8.

82. Piaggio, "Congenital Adrenal Hyperplasia," see note 78 above.

83. Baratz, Sharp, and Sanderg," Disorders of Sex Development," see note 62 above.

84. Lee et al., "Global Disorders of Sex Development," see note 40 above.

85. L.E. de Jesus, E.C. Costa, and S. Dekermacher, "Gender dysphoria and XX congenital adrenal hyperplasia: How frequent is it? Is male-sex rearing a good idea?" *Journal of Pediatric Surgery* 54, no. 11 (November 2019): 2421-7.

86. V. Pasterski et al., "Increased Cross-Gender Identification Independent of Gender Role Behavior in Girls with Congenital Adrenal Hyperplasia: Results from a Standardized Assessment of 4- to 11-Year-Old Children," *Archives of Sexual Behavior* 44, no. 5 (2015): 1363-75.

87. Lee et al., "Global Disorders of Sex Development," see note 40 above; C.P. Houk and P.A. Lee, "Approach to assigning gender in 46,XX congenital adrenal hyperplasia with male external genitalia: Replacing dogmatism with pragmatism," *Journal of Clinical Endocrinology and Metabolism* 95, no. 10 (2010): 4501-8; R. Gonzalez and B.M. Ludwikowski, "Should CAH in Females Be Classified as DSD?" *Frontiers in Pediatrics* 4 (2016): 48.

88. Societies for Pediatric Urology "Legislation," see note 36 above.

89. Stroumsa, "The state of transgender health care," see note 12 above.

90. Binet et al., "Should we question early feminizing genitoplasty," see note 76 above.

91. American Medical Association, "Michigan resolution: 013 (A-18): Opposing Surgical Sex Assignment of Infants with Differences of Sex Development," 2 May 2018, *https://www.ama-assn.org/system/files/2018-11/a18-resolutions.pdf,* p. 434; V. Pasterskiet et al., "Increased Cross-Gender Identification Independent of Gender Role Behavior in Girls with Congenital Adrenal Hyperplasia: Results from a Standardized Assessment of 4- to 11-Year-Old Children," Arch Sex Behav 44, no. 5 (Jul 2015): 1363-75. https://doi.org/10.1007/s10508-014-0385-0.

92. V. Pasterski et al., "Increased Cross-Gender Identification," see note 91 above.

93. J. Almasri et al., "Genital Reconstructive Surgery in Females With Congenital Adrenal Hyperplasia: A Systematic Review and Meta-Analysis," *Journal of Clinical Endocrinology and Metabolism* 103, no. 11 (2018): 4089-96.

94. A.R. Flores, J.L. Herman, G.J. Gates, and T.N.T. Brown, "How Many Adults Identify as Transgender in the

United States?" 2016, UCLA School of Law, Williams Law Institute, *https://williamsinstitute.law.ucla.edu/publications/trans-adults-united-states/.*

95. C. Dhejne et al., "Mental health and gender dysphoria: A review of the literature," *International Review of Psychiatry* 28 no. 1 (2016): 44-57.

96. S. Timmermans et al., "Gender destinies: Assigning gender in Disorders of Sex Development-Intersex clinics," *Sociology of Health and Illness* 41, no. 8 (2019): 1520-34.

97. Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," see note 8 above.

98. S. Timmermans et al., "Does patient-centered care change genital surgery decisions? The strategic use of clinical uncertainty in disorders of sex development clinics," *Journal of Health and Social Behavior* 59, no. 4 (2018): 520-35.

99. Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," see note 8 above.

100. Societies for Pediatric Urology and American Urological Association, "Pediatric Decision Making," see note 14 above.

101. Timmermans et al., "Does patient-centered care change genital surgery decisions?" see note 98 above.

102. Timmermans et al., "Gender destinies," see note 96 above.

103. Ibid.

104. Truog et al., "Microethics," see note 17 above.

105. Physicians for Human Rights, "Unnecessary Surgery on Intersex Children Must Stop," see note 8 above.

106. R. Chawla et al., "Utilization of a shared decision-making tool in a female infant with congenital adrenal hyperplasia and genital ambiguity," *Journal of Pediatric Endocrinology and Metabolism* 32, no. 6 (2019): 643-6.

107. Personal communication with Kathleen van Leeuwen, fourth co-author, ibid.