Intersex, DSD, and the Child's Well-Being: Changing Perceptions

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The medical management of people born with atypical genitalia has been in the focus of ethical and societal debates for several decades. This reflects both the advances in biological knowledge and significant sociocultural changes with regard to the concepts of sex and gender. In the 1950s, Money et al. [1], on the basis of their gender imprinting theory, recommended that children diagnosed with an intersex condition would benefit from a clear (surgical) assignment to a defined sex in a binary fashion in early infancy since gender identification would correlate with the visual appearance of their sexual organs. Information about this surgery and the underlying condition were not to be disclosed to the child. The idea was that gender assignment should not be questioned and that the affected child should not be confronted with the “anomaly.” However, Money et al. [2] already distinguished between different entities in the intersex spectrum and tried to associate this with a prognosis on the pubertal and adult sexual development and gender identity of the affected children. Toward the end of the 20th century, two main roads converged into a new approach. First, more and more affected adults accused the medical

community of being paternalistic and treating them inappropriately. Second, with growing expertise in how to characterize conditions at the molecular level, it was proposed that each person could be diagnosed and managed individually. This led to an internationally acclaimed consensus conference in 2005, which addressed the issues of nomenclature, classification, information policies, and management [3].

At that time, many medical colleagues thought that the term “intersex” should be abolished as it was assumed to be pejorative and not acceptable to the majority of people with conditions then gathered under the umbrella “disorders of sex development” or “DSD” [4] (later reworded as “*differences of sex development*”). The DSD classification aimed to abandon mythical phraseology (e.g., usage of words like hermaphroditism), refrain from stigmatization, and allow a strictly biological perception of already well-characterized entities with an openness to further evolution of biomedical understanding.

In the article on which we are commenting, Heino Meyer-Bahlburg addresses more recent developments in the field of DSD perception and management in this area [5]. Several legislatures and a recommendation of the Council of Europe [6] have issued a ban on medically unnecessary surgical procedures in minors to construct the genital appearance and function to appear as typically male or female. This follows demands from DSD advo-

cacy groups, who often use the term “intersex” for themselves proactively and associate with the LGBTIQI+ movement. Interestingly though, Meyer-Bahlburg uses the term “intersex” but now in conjunction with “somatic intersexuality,” challenging the DSD nomenclature despite its acceptance by the majority of people with such conditions [7]. He summarizes the findings of 10 surveys addressing the question of whether adults with DSD conditions would favor genital surgery in early childhood versus later in adolescence or adulthood, when their personal informed consent is possible. In summary, the majority of participants in these surveys answered that they retrospectively approved early surgery [8]. What can be concluded from this?

Before we discuss the ethics of this question, a few remarks about the science are to be discussed. Recent research indicates that the development of biological sex is a highly complex and variable phenomenon that is not limited to primary sexual binarity with the aim of reproductivity. Rather, maleness and femaleness are in fact multifaceted issues of gradual variations on many interacting levels: molecular, cellular, hormonal, bodily and corporeal development, psychology, family and other relationships, and society, affecting the material body, physical and psychological development, the emergence of behavioral traits, and social identity. Sex-differentiated gene expression occurs in almost all tissues and affects a broad range of body functions, far beyond sexual and reproductive functioning [9]. Moreover, stereotypical concepts of sex differentiation had and still have complex and significant outreach into society and culture, as well as from society and culture back into the understanding of the differential pathophysiology of diseases in human medicine.

The affirmation of scientific complexity underlying the *emergence* of two sexes also creates a new situation for discussion of the ethics of early surgical and hormonal gender reassignments. Meyer-Bahlburg points out that through continuously improved surgical techniques and increased knowledge about the psychosexual development in different underlying diagnoses, a treatment regimen has been established which leads to a satisfactory result for the *majority* of those operated on as children. Since the surveys reviewed indicate a majority of individuals with DSD who have meanwhile reached adulthood retrospectively favor early surgery, he concludes that “a mandatory delay of genital surgery to the age of consent disregards the preference of such patients and, thereby, constitutes a violation of medical ethics” [8]. We believe that this warrants a broad and serious discussion

since it may not be as clear as it seems. Which claims can the available evidence really establish? And which ethical conclusions can be justified on this basis?

Elective medical interventions in minors affect numerous (some of them legally protected) interests and ethically sensitive decisions, which are made in a complex web of individual, family-based, social, historical, legal, formal and informal norms, values, and beliefs. All seem however to agree that the child’s current and future best interests, the well-being of the child [10], should be at the center of any treatment decision. The medical-ethical ideal is, as philosopher Butler [11] has put it, “to do justice to someone,” i.e., to the individual person, which includes respecting the patient’s self-determination but which in a pediatric situation cannot be reduced to autonomy. In contrast to many other medical interventions in childhood, determining what will best serve the child’s interests and well-being in this situation is difficult. There are various reasons for this:

First of all, there is a severe lack of evidence on prospective surgical, psychosexual, and psychosocial outcomes, both on the individual and on the group levels. This is not only due to the rarity of DSD conditions and the multitude of different diagnoses summarized under this term but also mainly due to the lack of long-term prospective studies of different types of treatment outcomes (functional, esthetic, psychological, social, etc.) and treatment satisfaction, in the context of a changing society. We are dealing here with a glaring lacuna of long-term and context-sensitive medical and psychosocial knowledge. A further complicating factor is that the surgical techniques are constantly changing and will inevitably be further developed, with different approaches practiced in different institutions. All this makes it very difficult to compare retrospective study results to acquire broader evidence. To make matters worse, studies in the area of DSD often have only small case numbers, and therefore, the potential for recruitment bias is particularly important and must always be considered [12].

With regard to the surgical outcomes, it should also be noted that in many countries, genital surgeries performed in early childhood have been so common that there is little experience of how children develop without such an intervention. Parents and doctors often express fear of stigmatization, psychological problems, and impairment of sexual development, especially with regard to gender identity. But, there is also little evidence-based knowledge about whether and to what extent these fears are reasonable and which nonsurgical treatment offers could serve to counter stigmatization, psychosocial, and psychosexu-

al impairments. It is clear however that alternatives to surgery in the form of psycho-education and long-term psychological support are required. As Roen emphasizes: “When elective genital surgery is offered as a possible pathway, a credible and clearly supported nonsurgical pathway must be available as an alternative. Without an available alternative care pathway, (parents’) consent cannot be meaningfully given” [13].

Another aspect that has a strong influence on the evaluation of which treatment most closely corresponds to the current and future best interests of the individual child is social change, which in recent decades has led to strengthening of children’s and patient’s rights, bodily integrity, and protection against genital surgeries in many countries. It has also led to more open attitudes in societies toward people who do not identify with a binary sex or gender, either temporarily or long term. There is a clear trend toward greater acceptance and inclusion of individuals identifying as nonbinary and living outside the categories of “man” and “woman.” In some countries (e.g., Malta, Australia, Nepal, India, Germany), it is legally possible for people with DSD to register their official sex as intersex.

In his conclusions, Meyer-Bahlburg emphasizes (correctly, in our view) that increasing openness toward nonbinary identification is not universal and that it is impossible to estimate how this trend will develop. He relies on his clinical experience and points out that *most* children and adults with DSD would like to be assigned to either the male or female sex (even if they identify as nonbinary or inter* in private life). Meyer-Bahlburg states that the “intersex stigma” is still strong. But, how can we reinforce this with evidence? Unfortunately, the author does not name references here or specify which people are stigmatized, in which situations, and based on what aspects (behavior, genital features, appearance). The link between one’s own (alleged) desire for gender assignment and a high risk of stigmatization is often used as an argument for early surgeries. However, the inherent circularity of this argumentation is often overlooked.

One more thing seems crucially important. Refraining from surgery in early childhood *does not mean* that the child cannot be assigned to a gender (if the family and the child wishes to), and it does not mean either that genital surgeries may *never* be performed, for example, at a later point in time, or if it turns out that the child develops a high level of distress without surgery. The point is not to carry out any “preventive” surgeries but, whenever possible, to wait until the “affected” individual is able to (co-) decide. Ultimately, the task is to identify for each indi-

vidual case what the problem actually consists of, who has the problem, and what options there are for solving it. This may be the as yet unresolved conundrum of a biomedically diagnosed “DSD” and the “intersex” variability, which depends on individual self-perception in the eye of intended others.

As we have said, sex development occurs as a complex interaction of (epi-)genomes, hormones, other metabolites, cells, changes in morphology (during development in childhood, adolescence, and adulthood), psychological development, family relations at different stages of one’s biography, and evolving sociocultural gender patterns [5]. A better understanding of differentiation of a plurality of sexes and genders involves research on parts of this complex interactive pattern which can be meaningfully investigated but are partially but significantly interconnected.

Perhaps, this is the right moment to bring biology, medicine, and society into dialog on the issues of (non-) binarity, intersex, and DSD. New forms of multidisciplinary (involving many disciplines) and interdisciplinarity (involving scientists from many disciplines working together rather than in parallel) are needed. Truly interdisciplinary efforts can yield results that individuals and individual disciplines are unlikely to produce on their own.

Topics that push across new frontiers and provide research opportunities and potential for better understanding sex diversity, i.e., the multiple phenomena of diversity within the human sex-and-gender spectrum, include (1) research without the assumption of strict binarity in both the sex and gender systems, on all levels of development, expression, and influences. Binary structures can emerge and can be statistically detected, but research cannot assume that they represent natural kinds, and at a larger scale, they may lead to enormous variability. DSD as an entity within the variability of sex development can be a privileged research field where the potential of avoiding the dogma of binarity can be explored. (2) The complexity of interactions between cultural patterns of understanding (such as categories, ideas of ontological order) and biomedical research into sex development and clinical practices. (3) The history and contexts of clinical decisions and the values involved in clinical decision-making. This includes empirical ethics, normative ethics, law, and a better understanding of the complex temporality of the situation of decision-making, as well as limits to the ability to anticipate future personal needs in a society that is itself not static. (4) How the public sphere (including discourses in art, movies, literature, and the politico-legal

context) and the publicly visible performance of sex and gender (including social practices) influence the development of an individual's views and needs with regard to sexual identity and adjustment. (5) More comparative research on congenital and acquired symptoms of sex and gender variability would be desirable. (6) How the perspectives of affected persons, their families, and advocacy groups can be understood, both empirically and hermeneutically, in order to clarify the role of subjective experience and patient advocacy in good decision-making. (7) How stigma and discrimination work in reality. This can be addressed by serious inquiries into the multiple processes of discrimination on the basis of sex and gender diversity and development of strategies for inclusion that meet the needs and views of those who are to be included.

Some of these aspects have already been studied in other contexts but need to be revisited here under changing societal and biomedical conditions. Other aspects would need new forms of collaboration between scholars across the disciplines and ways to include patient advocacy groups. Having said this, we do not claim that no medical

decision-making is possible before all of this is achieved. But awareness of the complexity and of the limits of what we know about how patients view a treatment in the long run may prevent us from drawing conclusions too strictly and too soon.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

All the authors have equally contributed to the text.

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