

# THE IMPACT OF CULTURE ON SEX ASSIGNMENT AND GENDER DEVELOPMENT IN INTERSEX PATIENTS

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**ABSTRACT** The appearance of the external genitalia is the major determinant of the social sex, which is announced at or shortly after birth. In the absence of normal development of the external genitalia, definitive gender assignment and its announcement have to be postponed. While over the past 20 years the pathogenesis of most disorders causing abnormal development of the genitalia have been elucidated, our knowledge regarding the impact of these defects upon the psychosexual development is rather rudimentary. This information, however, is needed not only to establish criteria for correct sex assignment but also to design relevant outcome studies. Culture is an important part of the context in which decisions are made on sex assignment of patients with abnormalities of the external genitalia. Cultural differences in dealing with intersexuality and intersex individuals not only influences the patient's own psychosexual development but also medical decisions regarding sex assignment and consecutive management. There is evidence that attitudes concerning gender and sexuality, including the acceptance of intersexuality, differ significantly between various cultures. Thus cross-cultural studies might allow a new approach in dealing with intersexed persons, their families, and their social background, a most important aspect considering the recent discussions and criticisms of patients and individuals affected with intersex disorders.

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CULTURE IS AN IMPORTANT PART of the context in which decisions are made on sex assignment of patients with abnormalities of the external genitalia (Kessler 1990; Meyer-Bahlburg 1998). Cultural differences in dealing with intersex disorders might reasonably be thought to influence the process of psychosexual identification, commonly evaluated by considering gender identity, gender role, sexual orientation, and parenting. Gender identity is the awareness of oneself as male, female, or ambiguous; gender role is best explained by one's social behavior and role; and sexual orientation is defined by erotic responsiveness to one sex or the other. While these characteristics have commonalities across different cultures, it is unlikely that their expression would be totally invariant. Classically, however, cultural considerations have been less likely to be taken into overt consideration than clinical signs and symptoms, and many clinicians would probably deny being influenced by their own cultural background in the decision-making process. Across cultures, knowledge of the development of psychosexual identification of normal individuals and intersex patients is fragmentary. This is surprising given the relatively high incidence of abnormalities of the external genitalia: a conservative estimate from the medical literature suggests these abnormalities occur at the rate of 1 in 6,900 live births; the actual rate may be much higher, as suggested by others (Blackless et al. 2000; Castilla, Oriolo, and Lugarinho 1987).

During the last few years, discussion regarding the medical treatment of intersex individuals has increasingly evolved into criticism regarding the medical and surgical treatment of children born with genital abnormalities (Diamond 1982; Diamond and Sigmundson 1997; Dreger 1998; Fausto-Sterling 1996; Kessler 1998). This critical discussion had been initiated by adult patients who felt that the medical treatment of their condition had been inadequate and/or wrong (Chase 1998, 1999; Tomlin and Bergling 1999). Simultaneously, the interest in gender development has resulted in an increasing number of papers published by developmental psychologists and anthropologists comparing gender development in normal and intersexed infants (Dreger 1998; Kessler 1990, 1998).

Almost 10 years ago we performed and published the first comprehensive psychological and medical outcome study of patients with congenital adrenal hyperplasia, and our work has been followed by similar studies with similar results in Europe and the United States (Kuhnle, Bullinger, and Schwarz 1995; Kuhnle et al. 1993; Kuhnle et al. 1997; Slijper et al. 1998; Zucker et al. 1996). It is interesting to note that neither the medical papers—including our own—nor the anthropological research acknowledges or discusses each other's findings.

Our own critical reflection on the current policy of treating intersex infants had been initiated and stimulated while working in Southeast Asia. We had the opportunity to be involved in the management and to observe the outcome of patients with a variety of intersex disorders in Malaysia's largest children's hospital, affiliated with the National University of Malaysia. Our patients belong to the three main ethnic groups living in Malaysia, comprising 56 percent Malays,

33 percent Chinese, and 10.3 percent Indians, the remainder being other minorities. Virtually all Malays are Muslims; the Chinese are Buddhists or Taoists; and the majority of Indians are Hindus, with a smaller percentage being Muslims or Christians (Jamilah 1992).

Observing the development of intersex patients from different cultural backgrounds made it quite obvious to us that the current medical approach to intersexuality is guided more by cultural bias than by objective medical criteria. The idea that cultural background should guide medical decisions is nowhere generally accepted—neither among our medical colleagues in Europe and the United States nor among our Asian colleagues. Since the medical knowledge regarding the management of intersex patients is the same in Western and Southeast Asian medical teaching hospitals, we speculate that differences in outcome of intersex individuals are due to cultural differences and/or influences.

#### DEFINITION OF INTERSEXUALITY

What is so unusual about intersex genitalia or patients with intersex genitalia? In medical terms the definition of intersex genitalia is somewhat arbitrary. The terms *intersex*, *ambiguous*, or even *uncertain genitalia* reflect clinical uncertainty and medical ambiguity. These conditions include diagnoses like hypospadias and developmental problems of the external genitalia, virilized females, undervirilized males, and patients with gonadal tissue of both sexes. In some patients the origin of the abnormality is one of continuing defective hormone secretion or metabolism, whereas in others the problem reflects abnormal development of the sexual organs (Grumbach and Conte 1998).

The major difference from congenital malformation of other organ systems, such as abnormalities of the extremities, or facial abnormalities like cleft lip and palate, is that the appearance of the external genitalia is the major determinant of social sex, which is announced at or shortly after birth. In the absence of “normal” development of the external genitalia, definitive gender assignment and its announcement have to be postponed.

#### PATHOGENESIS OF INTERSEXUALITY

Over the past 20 years the pathogenesis of most intersex disorders has been elucidated, with the molecular defects causing the biochemical pathology identified. A number of clinical papers have outlined rapid and rational approaches to differential diagnoses, and diagnostic procedures are standardized and widely accepted. Criteria and recommendations for “correct” sex assignment include genetic, biochemical, and clinical findings; most endocrinologists and surgeons consider sex assignment a logical consequence of answers to these questions, and feel themselves appropriately guided by clinical judgement and laboratory results (Grumbach and Conte 1998).

Progress in terms of accuracy of the diagnosis has been tremendous: we can clearly distinguish whether the patient is male or female, with abnormal differentiation of the external genitalia, or a true hermaphrodite with both male and female gonadal tissue. Further pubertal development, as well as future fertility, can be predicted. The degree of virilization of the external genitalia is dependent not only on the amount of testosterone secreted, but also on the tissue sensitivity to androgens. It is accepted that androgens, which start to act in utero, also play a central role in determining male behavior, but both the time frame and the contribution of socialization remain essentially unknown (Berenbaum, Duck, and Bryk 2000; Collaer and Hines 1995; Ehrhardt, Epstein, and Money 1968).

### CRITERIA OF CORRECT SEX ASSIGNMENT

At this point, however, there arises the question of who decides what is the “correct” sex assignment, and who evaluates whether the assignment is appropriate. Put in a nutshell: what is *normal* in terms of sexuality? Cultural factors, education, and society play major roles in what we think is “normal” sexual behavior, which has changed markedly not only within the last 50 years. Studies on sexual behavior in the general population are rare, and studies in intersex patients are even rarer. In the clinical setting it is unclear if such studies are helpful, and indeed what sort of studies these might optimally be. Most studies on the outcome of intersexuality have used quality-of-life evaluations, which have the inherent problem that they frequently fail to detect deficits despite significantly impaired health status (Aaronson and Beckmann 1987; Bullinger and Hasford 1991; Bullinger and Pöpel 1988). We therefore have to ask what are the relevant questions and studies, and can cultural and clinical data be integrated and/or reconciled?

In previous centuries representatives of the law and the church, rather than physicians, observed the direction in which an intersex individual was developing, and then decided his or her appropriate place in society. At that time laws regarding public responsibilities of males and females differed significantly: for example, females could neither testify nor claim inheritance. Another possible problem was the fear of same sex marriages. Identification of the correct sex thus seemed to be very important. Final sex assignment was usually postponed until after puberty, and thus pubertal development as well as the patients’ own wishes could be taken into account (Dreger 1998; New and Kitzinger 1993; Wacke 1989).

In contrast, today physicians and parents decide on the best sex of an intersex child. The question asked is not so much what the individual is, but which gender is a better fit. The major criteria influencing decisions on sex assignment are functionality, future pubertal development, and fertility. Consequently, genetic male patients with phallic structures considered to be too small are raised as girls; whereas severely or completely virilized females with congenital adrenal hyperplasia are also raised as girls, with the main argument in the latter case being that future female fertility may be possible (Money 1994).

Furthermore, it is claimed that sex assignment should be within the first two years of life to allow the development of an unambiguous psychosexual identity. This hypothesis was developed and promulgated by psychologists from the John Hopkins Medical School in the 1960s and 1970s, and has found wide acceptance (Money 1974; Money, Hampson, and Hampson 1955). Unfortunately, this clinical decision tree was inappropriately extended from the notion that psychosexual identity itself develops during the first year or two of life to a corollary that external factors such as education and socialization were able to induce sexual identification. Together with the two major medical criteria of functionality and fertility, this extension has produced disastrous results in some cases of sex assignment and reassignment (Diamond 1982). The question therefore arises of whether this mindset may be part of the reason for the current unhappiness and dislocation of an increasing number of intersex individuals (AG gegen Gewalt 2001; Intersex Society of North America 1995).

The hypothesis of development of psychosexual identity put forward from Hopkins appeared very attractive at the time. Educational and social factors were considered the major determinants of human development, including personality and intelligence. Similarly consistent with these concepts were the statements of philosophers and feminists like Simone de Beauvoir and Monique Wittig, who claimed that “one is not *born* a woman,” a concept widely accepted not only among feminists (Beauvoir 1949; Wittig 1992).

The impact of improved biochemical diagnosis, early sex assignment and these psychological considerations has been that more intersex infants are being assigned to the female sex. This is well documented for true hermaphroditism and the virilizing forms of congenital adrenal hyperplasia (Krob, Braun, and Kuhnle 1994), and until very recently it was the clear medical, surgical, and psychological recommendation to raise boys with micropenis or severely undervirilized genitalia as girls (Money, Hampson, and Hampson 1955). The arguments for such an assignation were female fertility in XX individuals, and poor male sexual capacity in undervirilized boys. Has this treatment been successful? We do not know, since follow-up data are not available on these individuals. Besides some literature on outcome in virilizing congenital adrenal hyperplasia, information on the outcome individuals with severe hypospadias raised either as males or females is lacking (Aho et al. 2000; Federman 1987; Fryczkowski, Pradysz, and Krauze-Balwinska 1996; Kuhnle, Bullinger, and Schwarz 1995; Kuhnle et al. 1993; Lewis and Money 1983; Mulaikal, Migeon, and Rock 1987; Ringert, Hermanns, and Zoeller 1999; Schober 2001; Slijper et al. 1998; Tsinopoulou-Galli et al. 2002; Wisniewski et al. 2000; Woodhouse 1994; Zucker et al. 1996).

Patients with complete androgen insensitivity do not virilize at puberty, and these patients may constitute a different category, since their “female” identity is not questioned. In a recent report from the John Hopkins Medical Hospital, the first and so far only series of adult patients with complete androgen insensitivity was evaluated (Wisniewski et al. 2000). With the exception of one patient, all

lived in heterosexual relationships and a number had adopted children. Their quality of life was claimed to be high, with most patients expressing a high satisfaction in all aspects of life. The authors concluded not only that the general management of their patients had been satisfactory, but also that XY individuals have no difficulties to adjust to a life as a female. While this cannot be disputed as a generality, some individuals with androgen insensitivity syndrome may have problems in developing a sexual identity, be it male or female. To illustrate this we will present a German case of an XY individual with partial androgen insensitivity whose psychosexual identity was poorly developed, if at all (Tsinopoulou-Galli et al. 2002).

*Case 1:* During a family study of familial hypospadias due to a point mutation in the androgen receptor gene, we investigated and diagnosed the partial androgen insensitivity syndrome in a 30-year-old male who was born with severe penoscrotal hypospadias. The hypospadias had been corrected during childhood, and he developed gynecomastia during puberty, which was briefly treated without success with Danazolol; his micropenis was never treated. Semen analysis revealed the presence of sperm though the number and motility were significantly reduced.

The patient had had spontaneous ejaculations until the age of about 20 years, but he never had masturbated nor had sexual intercourse, though he claims to be attracted towards women. He denies any homosexual desire, and he never had any doubt of his maleness. Certainly he never wanted to be or to live as a woman. In his job as a professional he is well settled and recently built a house where he is—in his own words—“waiting for the right woman to marry.” On direct questioning he did not feel that he needed to take additional testosterone to increase his libido. His father later contacted us to voice his anxiety that his son might have a problem which needed intervention.

What can this case study tell us? The answer may be that to send questionnaires and to ask patients whether they are content with their life may not prove informative. All studies so far, including our own quality-of-life investigation of adult patients with congenital adrenal hyperplasia, produced patients who have been content with their life (Fryczkowski, Pradysz, and Krauze-Balwinska 1996; Kuhnle, Bullinger, and Schwarz 1995; Kuhnle et al. 1993; Lewis and Money 1983; Slijper et al. 1998; Wisniewski et al. 2000; Woodhouse 1994; Zucker et al. 1996). The conclusion drawn originally from these studies was that the treatment and management of these patients has been correct. We now believe that this is an unwarranted conclusion and that the only inference to be drawn from these studies is that the majority of patients are well adjusted and can somehow live with a handicap. These studies do *not* answer the question of whether there are other and/or better options for a patient's life.

Patients with complete androgen insensitivity syndrome are usually assigned to the female gender role. However, is it justified to study in such individuals

*female* libido, *female* sexual desire, and *female* psychosexual identity? The hypothesis of such studies is based upon the speculation that femaleness develops in a male in the absence of androgens, and as such is based on an absolutely unsubstantiated hypothesis that male development is the completion or the continuation of female development. Can we really test an hypothesis that *men* minus androgens develop into *women*?

Our patient with incomplete androgen receptor deficiency clearly contradicts this notion. He is a man with a decreased sexual activity and perhaps a disturbed “male” sexual identity, but certainly not a female nor even an incomplete female sexual identity. That he should live as a woman is beyond his imagination. His incomplete maleness does not make him in any way close to being a female. He is still a male, and whatever problems he has are related to his maleness. Using the wrong psychological test instruments is bound to give incorrect answers. Thus testing “female” signs in XY individuals is bound to give incorrect answers.

#### OUTCOME STUDIES IN PATIENTS WITH INTERSEXUALITY

The results of follow-up studies of intersexual individuals are almost unequivocal: intersexual individuals raised as males—even with severely malformed genitalia—are apparently able to lead satisfactory lives, with the majority of them married and claiming a fulfilled and happy married life (Aho et al. 2000; Reilly and Woodhouse 1989; Ringert, Hermanns, and Zoeller 1999; Wilson 1999; Woodhouse 1994). At first glance a similar scenario seems to be the case for virilized females with congenital adrenal hyperplasia. Quality-of-life studies show that most of these women have high levels of satisfaction and well-being and seem well settled in their female sex roles (Kuhnle, Bullinger, and Schwarz 1995; Kuhnle et al. 1993; Kuhnle et al. 1997; Slijper et al. 1998; Zucker et al. 1996). Nevertheless, the same studies show that a relatively large percentage of such women are neither married nor have children (Ehrhardt, Epstein, and Money 1968; Mulaikal, Migeon, and Rock 1987). This is of concern, given that fertility is one of the major arguments in assigning even severely virilized females to the female sex, and that fertility rates are considered a major indicator of good management and good therapeutic control (Federman 1987).

In this context a recent paper by Meyer-Bahlburg (1999) is of interest. Discussing the low fertility rates in congenital adrenal hyperplasia, this author points out that a number of adult patients with congenital adrenal hyperplasia actively avoid becoming pregnant despite good ovarian and vaginal function. He speculates that the reason might be lack of “maternalism,” introducing a dimension of intersexuality which to date has never been considered, let alone investigated.

The literature on outcomes for patients with congenital adrenal hyperplasia initially focused on medical therapy, primarily on the side effects of too much or too little glucocorticoid, and on the surgical outcomes of clitoral reduction and vaginal reconstruction (Alizai et al. 1999; Costa et al. 1997; Jones, Garcia, and

Klingensmith 1977; Klingensmith et al. 1977; Kuhnle 1996; Newman, Randolph, and Parson 1992). Ten years later studies focused on partnership and fertility rates. To draw the conclusion that lack of “maternalism” may be responsible for low fertility rates in women with congenital adrenal hyperplasia might be premature. “Maternalism” in experimental animals develops secondary to the hormonal changes during and not prior to pregnancy and parturition, and there is no particular reason to doubt that similar mechanisms are operative in humans, too (Bridges 1998; Bridges, Mann, and Coppeta 1999; Poindron, Levy, and Krebhiel 1988; Wang et al. 1995). Thus it is more likely that the low fertility rates in congenital adrenal hyperplasia may reflect problems surrounding partnership rather than a decreased wish for children.

### CULTURAL AND HISTORICAL CONSIDERATIONS

The apparent discrepancy between medical-psychological outcome data and the doubts voiced by patient support groups needs further investigation. As we have already discussed, part of this difference arises from the fact that quality-of-life studies used by most medical and psychological investigators do not reflect problems arising from a life with intersexuality, but rather the ability of intersex persons to develop coping mechanisms (Aaronson and Beckmann 1987; Bullinger and Hasford 1991; Bullinger and Pöpel 1988). To address the more important question of whether there might be better options in an individual's life, different studies are needed. The changing criteria by which to determine outcomes in congenital adrenal hyperplasia illustrate not only our current lack of established indices for optimal outcomes, but at an even more basic level the lack of criteria to decide on sex and gender assignment. To refine or recast the criteria for the management of the various forms of intersexuality, and to avoid coming to premature conclusions we need long-term observations on the lives of individual patients with various forms of intersexuality (Dreger 1999; Money 2000; Tsinopoulou-Galli et al. 2002).

In most societies the social and economic position of males and females differs significantly, and in all societies the male gender seems to offer more and better options in life (Ortner 1974, 1996). The authors' own personal experience extends from Europe and the United States to Malay Muslim, ethnic Indian, and Chinese societies in Malaysia, where women's roles differ in various ways. Furthermore, we have gotten the impression from working with families from different ethnic backgrounds that the acceptance of children with genital abnormalities differed significantly, too. Therefore, we speculate that our decisions regarding the treatment of intersexuality is influenced more by our own cultural background than by biological findings (or by the interpretation of biological data). In the following we would like to analyze briefly the cultural and ethnic differences of the three races living in Malaysia and present some data which in our opinion illustrates the different ways in which intersex patients are accepted. (Karim 1992, 1995).

The ethnic Malay women are Muslims. The Islamization of the Malay archipelago occurred about 1,000 years ago. The female role in religious and public life is somewhat comparable to the passive female role in the Christian church and Jewish temple. However, Malay Muslim women are entitled to inherit and control their own money, and with divorce, or at the death of her husband, the woman's fortune remains under her own control and thus can enable her to lead her own independent life. In addition, within a number of communities the original matrilineal society structures still exist, which among other things gives the women the exclusive right to own and inherit land. Through these traditions the independence and the economic power of Malay women can be substantial (Jamilah 1992; Karim 1995; Ong and Peletz 1995; Peletz 1996).

The condition of women is quite different in the ethnic Indian and Chinese communities, which both originate from patriarchal societies. The majority of Chinese and Indians migrated to Malaysia during the end of the 19th and the beginning of the 20th centuries as unskilled laborers trying to escape poverty in their own countries but also encouraged by the British colonial government. In neither culture or tradition were women able to inherit or control their own fortune. Partly due to religious taboos and partly due to the British policy of "divide and rule," all ethnic groups in Malaysia remained separated beyond independence in 1957, after which efforts were made to devise a common national system of education (Jamilah 1992).

The position of Chinese and Indian women within the Malaysian society has been less systematically studied than that of Malay women. However, both ethnic groups place high values on formal education, and girls from elite families in particular attended and profited from the English government-sponsored or Christian mission schools. This changed radically after 1970, when as a result of racial riots the New Economic Policy was launched, and Malay children (including girls) were systematically encouraged to attend schools of higher education. It was then that Malay women started to get engaged in politics and public life even before the better educated women of the other ethnic groups (Jamilah 1992; Karim 1992, 1995). While we were working in the two major medical schools of Malaysia, the National University of Malaysia and the University of Malaya, there were an equal number of Malay and Chinese professors, while the percentage of ethnic Indian university staff seemed to equal their percentage within the general population, with the proportion of female academic staff close to 50 percent.

Rapid economic development and growth, as well as the promotion of education, have been the determining factors in promoting the growing emancipation of women of all ethnic backgrounds. Still, differences remain. Among the Indian community girls usually mean a significant financial burden to the family, since depending on the social status of the family a significant dowry is expected, and to marry off several girls can be a financial disaster. In contrast, boys will increase the family's fortune (Ganguly-Scrase 2000).

Family and family relationships play a major role in both the private and public life of the ethnic Chinese communities. Family connections are extremely important and are the cornerstone of most successful businesses. Men are not only the head of the family and its fortune, but also the guarantor of its continuation. Family ties are thus crucial for wealth and family continuation, which is in addition reflected in the burial and death culture. Male members of the family are the only ones who can serve at the grave; without a male family member, burial cannot proceed, and the continuation of the family is threatened (Chan 2000).

It is beyond the scope of this paper to discuss the ways in which the patriarchal or matriarchal roots of the various cultures in Malaysia have influenced the current position of females within present-day Malaysian society (Peletz 1996). However, we got the impression that within the ethnic Chinese and Indian communities much more prestige was associated with the male role, underpinning the great importance given to male offspring in India and China itself (Tuljapurkar, Li, and Feldman 1995). While we were working with different ethnic groups, it was never difficult to convince a Muslim family to assign a severely virilized girl or an undervirilized boy to the female gender. This was not the case for Chinese and Indian families, who on several occasions took off with their ambiguously born child when female sex assignment (or reassignment) was suggested.

Even though it seemed easier to work with Muslim parents and their newborn children with ambiguous genitalia, this does not mean that these families easily accept the diagnosis (Abdullah et al. 1991; Taha and Magbol 1995). Compliance with therapy was often very poor, and as a consequence the children virilized before or around puberty, which was probably taken by the parents as a sign that the medical diagnosis was not correct. This might be the explanation that changes at puberty caused surprisingly less confusion than the original intersexuality at birth, as illustrated by the following case report. Whether gender role changes that have been observed and reported by anthropologists to occur within certain Indonesian societies—a culture closely related to the Malaysian society—are comparable with our observations cannot be decided using the available data base. But it is of great interest for further gender role studies that within certain Southeast Asian cultures, gender roles are flexible and allow certain individuals to change from the male to the female gender role and vice versa, almost whenever it is felt to be convenient (Röttger-Rössler 1994, 1997).

*Case 2:* Fatimah was born as a severely virilized girl and diagnosed correctly to have congenital adrenal hyperplasia at birth. Sex assignment was female, and treatment with steroids was started. Obviously due to poor compliance with medication the child continued to virilize and underwent male puberty. At primary school she started to socialize with boys and was accepted with no apparent problems into the male society as a peer, not as a tomboy who likes to play

with boys. She prayed with the males in the mosque, was circumcised, and was accepted into the male role in religious life at the appropriate age. His maleness was never questioned by his peers. When we saw him at the age of 12 years, his clothing and the way he moved and behaved was without any doubt masculine. Why did we see him at all at the age of 12? He had applied for an identity card, and after the officer saw him he refused to issue one with his given name Fatimah. Until that point neither the patient nor his parents had felt that they needed medical help.

With the assistance of the religious leader of the community, Fatimah's original female sex assignment was changed to male. The celebration of his circumcision was arranged by the family even before corrective genital surgery could be performed.

### DETERMINANTS OF SEX ROLE BEHAVIOR

Patients like this lead to the question of when in life psychosexual identity develops and sex assignment is irreversible. Is it a major concern if there has been a mistake in sex assignment at birth? Is socialization a major factor for the development of psychosexual identity, and thus can parents educate or doctors treat children into the opposite *biologic* sex (Diamond and Sigmundson 1997; Money, Devore, and Norman 1986)?

At this point it seems important to emphasize again that even though we know a lot about the biology of normal and defective sexual development, we know little about how and when normal psychosexual identity develops, and less about what are the essential factors in such development. In short, we do not know at this point how to determine in early life *psychologic* sex. This is not due to lack of interest: research on gender-related development has been increasingly popular (Giesen 2000; Ruble and Martin 1998). We feel that one of the crucial topics within this literature is the unresolved debate on nature versus nurture and the question of whether and how early children learn gender roles versus the influence of genes and hormones on the development of behavior, gender identity, and sexual orientation. Within the current medical literature on the management of intersexuality, it seems that the nurture hypothesis as established by John Money and his followers from the Johns Hopkins Medical School is most widely accepted and has entered the medical textbooks (Lewis and Money 1983; Money 1994).

Earlier in this paper we criticized the consequences of using Money's developmental hypothesis in treating intersex children—a criticism that has been also voiced by others (Garrels 1998; Meyer-Bahlburg 1993, 1998). In our opinion, however, the major criticism does not arise from an extensive interpretation and use of the nurture hypothesis, but more from the inadequate interpretation and use of biological data. Money and his group were the first to study the influence of defective hormone secretion on the development of psychosexual identity

(Ehrhardt, Epstein, and Money 1968; Money, Schwartz, and Lewis 1984; Money, Devore, and Norman 1986). They found that androgen excess resulted not only in physical but also in psychological virilization of genetic females, while defective androgen secretion resulted in defective genital and psychological male development, interpreted as “feminizing” defects (Money 1974). Still, their treatment recommendations were dependent more on social adaptation and fertility than on the potential influence of androgenic substances on brain and behavior development (Lewis and Money 1983). Certainly applying a nurture approach to managing children with intersexuality does not include patients’ own decisions regarding gender role or timing of genital corrective surgery.

However, increasingly persons with intersexuality claim that they would have preferred to be included in the decision-making process regarding their sex assignment, as well as in decisions regarding definitive genital operations, some going as far as claiming a third sex category (AG gegen Gewalt 2001). In Western cultures a third sex category does not exist, and it seems to us impossible to advise parents how to raise a child beyond being male or female; it might, however, be possible to delay genital operations until the patients can decide for themselves. In some non-Western cultures third sex categories do exist, but our knowledge of such lifestyles is minimal. Intersex individuals in India live separately, as Hijaras, but it seems doubtful that this means fewer rather than more restrictions (Cohen 1995; Nanda 1990). None of our Asian patients, even though some had severely ambiguous genitalia, lived in this category.

How far can cultural factors and/or the wish for social advantage influence the decision to live as a member of a particular sex? To what extent is it the decision of the individual, and to what extent that of society, to accept a person in a particular sex role? What are the mechanisms which make us acceptable to the other members of society in our own sex role, and is there a timeframe which limits at what time in life we can finally be accepted in this position?

With the third case report, we would like to illustrate that culture and society can remain flexible and allow gender roles to shift from one sex to the other.

*Case 3:* Diana was referred at the age of 12 years to the Children’s Hospital in Kuala Lumpur from Sarawak, East Malaysia. She had been born as the fifth child to an ethnic Chinese family and was assigned female. The family already had two boys and two girls. At about the age of one year, the mother became increasingly worried about the looks of the external genitalia of her youngest daughter and brought her to a local general practitioner. This medical man agreed that the external genitalia of this girl were ambiguous, but he had no way of telling what the correct sex should be. His advice was to come back at 12 years of age, and until then wait and see what develops. That was the reason why the family sought advice at this point in time.

According to the medical history, the child had had no severe illness and developed normally. She attended a girls’ school and was a straight-A student. During school she wore a school uniform, but as soon as she returned home

she changed into loose T-shirts and wide shorts, an outfit never worn by Chinese girls in Malaysia. In addition her hair cut was extremely short and styled like a boy's, also culturally unacceptable for girls. However, this is tolerated by the family and her friends. She has a number of very good girl friends and is socially well integrated; she does not mix with boys, a behavior which is normal for a girl of her age in this culture and setting.

When she arrived at the ward, her manners, her body language, and her outfit were such that everybody considered her first as a boy. Her body habitus was rather muscular, with sparse body hair which was normal for age and race, and she had no beard and no change of voice yet. Her genitalia can be described as follows: a phallus of about 5 cm in length with significant erectile tissue, severe hypospadias with a single penoscrotal opening, severe chordee, a bifid scrotum with two somewhat soft testicles palpable with a volume of app. 6 ml each. Biochemically we could exclude a 5-alpha reductase deficiency, as well as an androgen receptor defect. The most likely diagnosis was partial gonadal dysgenesis.

On her first psychological examination by a child psychiatrist, she claimed to be a girl and asked for corrective surgery. However, this intention soon changed, and she herself as well as both parents requested that her social sex should be changed into the male gender, which was readily accepted by all medical doctors involved. He had corrective surgery and returned to Sarawak. Unfortunately, his urethra developed strictures and multiple fistulas, and he had to be repeatedly hospitalized. Due to his rather distant home in a small town in Sarawak, no continuous psychological help could be offered. His school performance dropped and finally he was lost for follow-up.

Despite the unsatisfactory medical follow-up of this patient, the ease with which he had switched during his life from one gender to another already prior to his medical treatment imposes the question of who influences gender development in normal but also in intersex individuals. Is culture or society imposing a certain gender role, or do individuals shape their own gender roles? The few available case reports, including our own, seem to indicate that intersex individuals find their own gender independent and maybe even undisturbed by external factors (Meyer-Bahlburg et al. 1996; Tomlin and Bergling 1999).

Similar observations have been described first by Imperato-McGinley, Peterson, and Gautier (1979), and later by Herdt (1990) in isolated and/or indigenous populations with intersexuality due to 5-alpha reductase deficiency, as an example. Affected individuals are reared in these societies as girls, but after virilizing at puberty they adopt a male sexual identity and are accepted within male society, seemingly without any problems. Several similar isolated cases have subsequently been reported from all parts of the world; in all such cases it seems that male identity develops around puberty, and most individuals continue their adult life as males (Imperato-McGinley et al. 1980; Wilson, Griffin, and Russell 1993). As a consequence, a number of patients with either 5-alpha reductase deficiency or partial androgen insensitivity syndrome are now reared as boys, even though

their genitalia at birth and during childhood were severely undervirilized (Diamond, Binstock, and Kohl 1996; Hiort 1999). If this indicates that the appearance of the external genitalia during childhood plays a minor role in the development of gender identity, it might also mean that gender identity is relatively independent of education and socialization.

There are, however, some reports in the ethnological literature which seem to contradict this theory, describing traditions in which parents educate a girl as a boy in case there is no male child in the family (Gremaux 1994; Wikan 1977). Unfortunately, none of these reports has ever been accompanied by medical examinations or hormonal data, and thus we do not know whether such children were sexually ambiguous from the outset, and whether perhaps these traditions developed to overcome a perceived difficult situation for the family. Intersex children are born in all societies, and each society has to have ways to deal with them. Our knowledge of the mechanisms involved is remarkably poor; there are very few analyses in Western societies, and even fewer in most other parts of the world. Most written documents are single case reports, or personal autobiographies without medical documentation.

### CONCLUSION

At this point we know very little about the complex influences of culture, society, and biology, as reflected by hormones but also genes and gonads upon the development of the psychosexual identity. Most factors contributing to and/or disturbing the development of a stable gender and sexual identity are unknown. At least in certain cultures, sex and gender role behavior can develop relatively independent of sex assignment and sex-specific education. The acceptance of intersex individuals by society is remarkable, although the signals and mechanisms by which this is achieved are unknown.

Before new studies can be designed, one has to go back and reevaluate the various forms of intersexuality. The answer may lie in the detailed and careful long-term analysis of life histories of intersexual persons, and in identification of the factors necessary for the development of a male or female psychosexual identity. We agree with John Money (2000), who stated that these life histories can “provide the opportunity to tease out the conflation of factors that may be responsible for a given outcome.”

Cross-cultural studies certainly may be helpful to resolve some of our difficulties, but they are hard to conduct. They may, however, be the key to our understanding of “what makes a man a man” (McLaren 1990) and a “woman a woman,” since certainly women cannot be taken as the result of deficient male development.

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