

Experiment of Nurture: Ablatio Penis at 2 Months, Sex Reassignment at 7 Months, and a Psychosexual Follow-up in Young Adulthood

Susan J. Bradley, MD*; Gillian D. Oliver, MD‡; Avinoam B. Chernick, MD§; and Kenneth J. Zucker, PhD||

ABSTRACT. Guidelines of psychosexual management for infants born with physical intersex conditions are intended to assist physicians and parents in making decisions about sex of assignment and rearing including the following: 1) sex assignment should be to the gender that carries the best prognosis for good reproductive function, good sexual function, normal-looking external genitalia and physical appearance, and a stable gender identity; 2) the decision regarding sex assignment should be made as early as possible, preferably during the newborn period, with an upper age limit for reversal of an initial sex assignment no later than 18 to 24 months; and 3) there should be minimal uncertainty and ambiguity on the part of parents and professionals regarding the final decision about sex assignment and rearing.

J. Money used these guidelines in a case of a biologically normal male infant (one of a pair of monozygotic twins) whose penis was accidentally ablated during a circumcision at the age of 7 months. The decision to reassign the infant boy to the female sex and to rear him as a girl was made at 17 months, with surgical castration and initial genital reconstruction occurring at 21 months.

Money reported follow-up data on this child through the age of 9 years. Although the girl was described as having many "tomboyish" behavioral traits, a female gender identity had apparently differentiated. Thus, it was concluded that gender identity is sufficiently incompletely differentiated at birth as to permit successful assignment of a genetic male as a girl, in keeping with the experiences of rearing.

Subsequent follow-up by other investigators reported that by early adolescence the patient had rejected the female identity and began to live as a male at the age of 14 years. In adulthood, the patient recalled that he had never felt comfortable as a girl, and his mother reported similar recollections. At age 25, the patient married a woman and adopted her children. The patient reported exclusive sexual attraction to females.

The present case report is a long-term psychosexual follow-up on a second case of ablatio penis in a 46 XY male. During an electrocautery circumcision at the age of 2 months, the patient sustained a burn of the skin of the

entire penile shaft, and the penis eventually sloughed off. At age 7 months, the remainder of the penis and the testes were removed. By age 7 months, if not earlier, the decision was made to reassign the patient as a female and to raise the infant as a girl.

The patient was interviewed on two occasions: at 16 years and twice while in the hospital for additional surgery at 26 years of age. At ages 16 and 26, the patient was living socially as a woman and denied any uncertainty about being a female. During childhood, the patient recalled that she self-identified as a "tomboy" and enjoyed stereotypically masculine toys and games; however, the patient also recalled that her favorite playmates were usually girls and that her best friend was always a girl.

When seen at age 16, the patient had been admitted to the hospital for vaginoplasty. At that time, she wished to proceed with the further repair of her genitalia to make them suitable for sexual intercourse with males. At age 26, the patient returned to the hospital for further vaginoplasty.

Regarding the patient's sexual orientation, she was attracted predominantly to women in fantasy, but had had sexual experiences with both women and men. At the time of the second surgery, she was in a relationship with a man and wished to be able to have intercourse. The patient's self-described sexual identity was "bisexual." After surgery at age 26 years, the patient developed a rectovaginal fistula. Within a few months of the surgery, the patient and her male partner separated for reasons other than the patient's physical problems. The patient subsequently began living with a new partner, a woman, in a lesbian relationship.

The psychosexual development of our patient was both similar to and different from the patient described earlier. Our patient differentiated a female gender identity; in contrast, the other patient had adopted a male gender identity after experiencing intense discomfort living as a female, apparently around the beginning of adolescence. At the time of interview at age 26, our patient was living with a man, but they subsequently separated and she began a new relationship with a woman; the other patient was married to a woman. Our patient had a "bisexual" sexual identity; the other patient had a "heterosexual" sexual identity. The patients were similar in that they had a childhood history of "tomboyism." Our patient was predominantly sexually attracted to women; the other patient was exclusively sexually attracted to women. Our patient had sexual experiences with both women and men; the other patient had sexual experiences only with women.

The most plausible explanation of our patient's differentiation of a female gender identity is that sex of rearing as a female, beginning at around age 7 months, overrode any putative influences of a normal prenatal masculine sexual biology.

Because cases of ablatio penis in infancy are so rare and long-term follow-up data are scant, it is obviously

From the *Department of Psychiatry and ‡Division of Gynaecology, Hospital for Sick Children, Toronto, Ontario, Canada; §Private Practice, London, Ontario, Canada; and the ||Child and Adolescent Gender Identity Clinic, Child and Family Studies Centre, Clarke Institute of Psychiatry, Toronto, Ontario, Canada.

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Reprint requests to (S.J.B.) Child and Family Studies Centre, Clarke Institute of Psychiatry, 250 College St, Toronto, Ontario M5T 1R8 Canada.

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impossible to know whether our patient or the previous case would be more typical of the psychosexual outcome in a larger sample of such individuals. However, our case suggests that it is possible for a female gender identity to differentiate in a biologically “normal” genetic male. At present, however, the clinical literature is deeply divided on the best way to manage cases of traumatic loss of the penis during infancy. Further study is clearly required to decide on the optimal model of psychosocial and psychosexual management for affected individuals. *Pediatrics* 1998;102(1). URL: <http://www.pediatrics.org/cgi/content/full/102/1/e9>; *ablatio penis, sex reassignment, gender identity, sexual orientation, physical intersex conditions*.

ABBREVIATION. CAH, congenital adrenal hyperplasia.

In the 1950s, Money and colleagues^{1,2} developed guidelines of psychosexual management for infants born with physical intersex conditions. They were intended to assist physicians, other health professionals, and parents in making decisions about sex assignment and rearing. These guidelines included the following: 1) sex assignment should be to the gender that carries the best prognosis for good reproductive function (if attainable at all), good sexual function, normal-looking external genitalia and physical appearance, and a stable gender identity (sense of self as a boy or a girl), all of which would putatively foster a healthy psychosocial adaptation^{3,4}; 2) the decision regarding sex assignment should be made as early as possible, preferably during the newborn period, with an upper age limit for reversal of an initial sex assignment no later than 18 to 24 months⁵; 3) there should be minimal uncertainty and ambiguity on the part of parents and professionals regarding the final decision about sex assignment and subsequent sex of rearing.^{5,6} As noted by Meyer-Bahlburg,⁴ these guidelines have informed the standard medical and psychosocial care of infants born with physical intersex conditions for several decades in the United States and other countries.

Follow-up studies of infants with physical intersex conditions showed that sex of rearing, in comparison with the various parameters that constitute biologic sex, was the best predictor of subsequent gender identity formation. For example, most genetic females with congenital adrenal hyperplasia (CAH) reared as girls developed a female gender identity,^{7,8} even though they had been exposed prenatally to increased levels of androgens^{9,10} (although subsequently controlled postnatally by glucocorticoid replacement therapy)¹¹ and were born with masculinized genitalia (although surgically feminized postnatally, usually in the first few years of life).¹² Parents reported that they were able to rear their children as girls with little reservation or ambivalence.⁷

There have, however, also been cases of gender change from female to male later in life in CAH genetic females, which have been associated with delayed decisions about sex assignment, chronic ambiguity about sex of rearing, inconsistent adherence to glucocorticoid replacement therapy, and lack of

surgical feminization of the external genitalia.¹³ These findings underscore the importance of both psychosocial and medical factors in the clinical care of infants born with physical intersex conditions.

Money^{14,15} applied the guidelines of psychosexual management for intersex children to a case of a biologically normal male infant (one of a pair of monozygotic twins) whose penis was accidentally ablated (flush with the abdominal wall) during a circumcision by electrocautery at 7 months of age. It then necrosed and sloughed off. The decision to reassign the infant boy to the female sex and to rear him as a girl was made when he was 17 months of age, with surgical castration and initial genital reconstruction performed at 21 months of age, which fall at the upper bound of the age range recommended for sex assignment (or reassignment) of infants born with physical intersex conditions.⁵

Money¹⁵ reported follow-up data on this child through 9 years of age, at which time the patient was described as having many “tomboyish traits, such as abundant physical energy, a high level of activity . . . and being often the dominant one in a girl’s group.”¹⁵ Money reported, however, that a female gender identity had apparently differentiated: “Her behavior is so normally that of an active little girl, and so clearly different by contrast from the boyish ways of her twin brother, that it offers nothing to stimulate one’s conjectures.”¹⁵ Thus, Money concluded that “gender identity is sufficiently incompletely differentiated at birth as to permit successful assignment of a genetic male as a girl . . . and differentiates in keeping with the experiences of rearing.”¹⁵ When Money first reported the case, it received widespread media attention¹⁶ and was noted in many pediatric, psychology, and sociology textbooks as a powerful proof of the importance of environmental influences on gender identity formation.¹⁷

Subsequently, however, Diamond¹⁷ reported the further course of events for this patient. By early adolescence, the patient had rejected the female identity and, at 14 years of age, began to live as a male.¹⁸ Indeed, when interviewed in his early 30s, the patient’s recall of his childhood gender development was that he had never felt comfortable as a girl, and his mother reported similar recollections. At age 14, the patient received a mastectomy and began testosterone replacement therapy; surgical procedures for phallus construction were performed at 15 and 16 years of age. At 25 years of age, the patient married a woman several years his senior and adopted her children. The patient reported exclusive sexual attraction to females.¹⁸

The long-term psychosexual outcome of this patient, which also received recent widespread media attention,^{19–22} has been used as evidence against the importance of sex of rearing for gender identity formation and also as a general critique of the guidelines of psychosexual management that have been used in the care of infants with physical intersex conditions.^{18,23}

Unfortunately, the factors that resulted in the long-term outcome for the case remain unclear. On the one hand, the eventual adoption of a male gender

identity and the emergence of sexual feelings toward women could be viewed as primarily attributable to the influence of a normal male sexual biology in utero.^{18,23,24} On the other hand, the failure of the female gender identity to maintain itself by adolescence could be attributed to psychosocial factors, such as parental ambivalence regarding the initial decision to reassign the infant as a girl. Ultimately, all one can conclude is that the experiment of nurture eventually failed, but why it did cannot be determined.

The present case report is a long-term psychosexual follow-up on a second case of ablatio penis, alluded to by Money¹⁵ but reported here for the first time.

CASE REPORT

The patient is a chromosomal 46 XY male. During an electrocautery circumcision at 2 months of age, the patient sustained a burn of the skin of the entire penile shaft, and the penis eventually sloughed off. Consequently, the patient was unable to void through the urethra, resulting in a suprapubic cystostoma. The patient was hospitalized subsequently for care of the surgical complications, and at 7 months of age was referred to Johns Hopkins Hospital (by ABC, the mother's obstetrician and the physician who had delivered the patient), where the remainder of the penis and the testes were removed. The suprapubic cystostoma was eventually closed, and the patient was then able to void through the urethra. Sometime between the circumcision accident and the hospital admission, the decision was made to reassign the patient as a female and to raise the baby as a girl. This was recognized formally at the time the patient was admitted to Johns Hopkins, because the infant was identified in hospital records as female.

The patient was interviewed on two occasions by a psychiatrist (SJB): at 16 years of age and twice while in hospital for additional surgery at 26 years of age. When the patient was 16, her mother also was interviewed. At age 26, the patient also was evaluated independently by a gynecologist (GDO) and then, after surgical complications (see below), by ABC several months later.

At 16 and 26 years of age, the patient was living socially as a woman. She denied any uncertainty about being female from as far back as she could remember and did not report any dysphoric feelings about being a woman. Indeed, three of the present authors who interviewed the patient (SJB, GDO, ABC) were in agreement that she was comfortable living as a woman. She recalled that during childhood, however, she self-identified as a "tomboy" and enjoyed stereotypically masculine toys and games. On the other hand, the patient had not engaged in cross-dressing or other forms of passing as a boy, and she recalled that her favorite playmates usually were girls and that her best friend was always a girl.

The patient's perception was that her mother was a "matter-of-fact" individual who, when told that she needed to rear her child as a female, simply did so. The patient indicated that in this respect she is much like her mother and, despite numerous adversities, has decided that she simply has to get on with life and do whatever she can rather than worry or obsess about issues.

Before the patient's birth, the parents were having long-standing marital difficulties and were seen for counseling by ABC. The patient's conception was motivated, in part, by the parent's attempt to salvage their marriage; however, the parents eventually divorced when the patient was approximately 3 to 4 years of age, in part because of the father's alcoholism but also because of the father having had a greater difficulty than the mother in dealing with the "loss" of his son. After a few years, her mother remarried and the patient developed a positive relationship with her stepfather, although there were some concerns about his drinking and his relationship with her mother.

Two months before the patient's 11th birthday, she was started on feminizing hormonal therapy (Premarin), which has continued to the present time. The patient was told about the circumcision and penile ablation when she was 12 years of age because she had begun asking her mother whether there was something wrong with her. During the interview with the mother when the patient

was 16, she reported that the penile stub, which had been left after the initial surgery, became erect when the patient showered or swam, and that this caused some difficulties dressing her in bathing suits. At 16 years of age, the patient reported that she was aware that her genitalia looked neither exactly like those of other girls nor like those of a boy.

When seen at age 16, the patient had been admitted to hospital for vaginoplasty. At that time, she denied any concerns about her gender identity and clearly wished to proceed with the repair of her genitalia to make them suitable for sexual intercourse with males.

At 26 years of age, the patient returned to hospital for additional vaginoplasty. She reported difficulty with intercourse after her earlier surgery because the vaginal opening was too small. At the time of the second surgery, she was in a relationship with a man and wished to be able to have intercourse. She also reported three significant sexual relationships with women. The patient noted that she found women more physically attractive than men, especially when naked. She found male genitalia "funny"; however, she found men sexually attractive if they were clothed or in underwear. In addition to the three relationships with women, the patient reported three sexual relationships with men. She reported better relationships with men in terms of sharing interests (eg, in her occupation) and a sense of openness with them. She felt, however, that she got along better sexually with women. In the relationship with her current male partner, she reported that she had tried to scare him off, apparently out of a sense that he would reject her when he knew her. Despite these rebuffs on her part, they appeared to be getting along well and she had begun to realize that he was, in fact, accepting of her and liked her for who she was.

Physically, the patient presented as a tall (175 cm), thin female, casually dressed but readily perceived as a woman. On examination at age 26 (performed by GDO), her breast development was Tanner stage V with a B size cup. Abdominal and pubic examination revealed a mons with a midline deficit consistent with her two previous reconstructive surgeries. The external genitalia showed prominent, redundant, bilateral rugated labia majora with a positive cremasteric reflex. The remains of the penile glans were consistent in size with a normal clitoris but deviated to the right. It was positive for sensation. She had a palpable, thick chord of erectile tissue running from the glans over the pubic symphysis, remnants of the corpus spongiosum. She specifically complained of engorgement of this area with sexual arousal. She had a 2-cm mass on the anterior aspect of the left labia majora which she found irritating; it was later determined to be the remnants of the scrotal sac. The vaginal introitus had a reasonable opening, acceptable for intercourse; however, there was a stenotic band on the right anterior margin which she complained made intercourse painful. The vaginal canal accepted two fingers with good length, but deviated 30 degrees to the patient's left and reportedly made penile-vaginal intercourse impossible. She reported arousal with stimulation of the reconstructed clitoris and her breasts.

The patient denied ever feeling that she had wanted to be a male and saw many advantages to being female; however, she readily acknowledged masculine interests, consistent with her occupation in a "blue collar" job practiced almost exclusively by men. She did not report a desire to become a parent. Despite her masculine occupational and recreational interests, she related to the interviewer as a female, discussing her dilemmas around her current relationship. She reported that previously she used to dislike her nose and thought that she was too skinny and too tall, but now feels that the only part of her body that bothers her are her genitals. This was related to their interfering with her being able to have intercourse. Her overall lifetime score on the Kinsey scale²⁵ (a 7-point rating scale, where 0 = exclusive heterosexuality and 6 = exclusive homosexuality) in fantasy was a 5 (predominant sexual attraction to women) and in behavior was a 3 (equivalent amounts of sexual experience with men and women). The patient's self-described sexual identity was "bisexual."

Unfortunately, after the most recent surgery, the patient developed a recto-vaginal fistula. Within a few months of the surgery, the patient and her male partner separated for reasons other than the patient's physical problems. The patient's post-surgery complications have been monitored by ABC and she reported to him that she was currently living with a new partner, a woman, in a lesbian relationship.

DISCUSSION

The psychosexual development of our patient was both similar to and different from the patient described earlier.^{15,17,18} Our patient differentiated a female gender identity, with no evidence of gender dysphoria in childhood, adolescence, and adulthood; in contrast, the other patient had adopted a male gender identity after experiencing intense discomfort living as a female, apparently from approximately the beginning of adolescence,¹⁷ if not earlier.^{18,22} At the time of the interview at age 26, our patient was living with a man, but they subsequently separated and she began a new relationship with a woman. The other patient was married to a woman. Our patient had a bisexual sexual identity; the other patient had a heterosexual sexual identity. The patients were similar in that they had a childhood history of tomboy-like behavior, including a high activity level and a preference for stereotypically masculine toys. However, our patient also recalled healthy friendships with other girls, compared with the other patient, who was apparently rejected and socially ostracized by other girls.¹⁷ Our patient was predominantly sexually attracted to women; the other patient was exclusively sexually attracted to women. Our patient had sexual experiences with both women and men; the other patient had sexual experiences only with women.

The most plausible explanation of our patient's differentiation of a female gender identity is that sex of rearing as a female, beginning at approximately 7 months of age, overrode any putative influences of a normal prenatal masculine sexual biology. Although it is not possible to state with precision what constituted our patient's sex of rearing as a girl, it clearly included her parents agreeing to the sex reassignment decision (although this was easier for the mother than for the father), the adoption of a stereotypically female name, and the patient being perceived as a girl by significant others in her social environment. In this case, then, the experiment of nurture was successful regarding female gender identity differentiation.

There are two likely reasons, perhaps related, why the gender identity development of our patient differentiated as a female, whereas it did not (at least in the long run) in the previous case. First, in our case, the decision to reassign the patient to the female sex occurred approximately between 2 and 7 months of age; in the other case, it occurred only at 17 months, and surgical castration and vaginoplasty were performed at 21 months. Second, it is possible that the parents of our patient, particularly the mother, had less ambivalence about the decision than the parents of the other patient, perhaps because the decision was made earlier in the patient's life.

It is, however, of importance that our patient had a strong history of behavioral masculinity during childhood and a predominance of sexual attraction to females in fantasy. Girls with CAH also display masculine gender role behavior^{7,26} and, in adulthood, are more likely to be bisexual or homosexual than are female controls.^{8,27,28}

Some would argue, therefore, that both the behavioral masculinity in childhood and the predominance of sexual attraction to females in adulthood are related to our patient's normal masculine sexual biology in utero, particularly with regard to prenatal androgenization of the central nervous system.^{29,30} Perhaps, therefore, these two components of psychosexual differentiation—gender role and sexual orientation—are more strongly influenced by biologic factors than is gender identity formation. This conjecture is consistent with experimental research on lower animals in which, for example, it is well established that female fetuses exposed to increased levels of androgen exhibit a shift toward male-typical patterns of gender role behavior and sex-of-partner preference.^{31–33} There is, however, no comparable animal analogue for human gender identity formation, given its subjective, phenomenologic nature.

Nonetheless, it also is important to note that our patient desired, at least at the time of our evaluation, to pursue a sexual relationship with a man; indeed, her primary reason to seek additional vaginoplasty in adulthood was to enhance the sexual aspect of that relationship. However, during the course of our interviews with the patient at age 26, she switched from living with a man to living with a woman, suggesting that her partner preferences remain in flux, and the long-term nature of her interpersonal sexual relations is uncertain.

Because cases of ablatio penis in infancy are so rare, and long-term follow-up data are scant, it is obviously impossible to know whether our patient or the previous case would be more typical of the psychosexual outcome in a larger sample of such individuals. However, our case suggests that it is possible for a female gender identity to differentiate in a biologically normal genetic male, which supports the original conclusion of Money et al⁵ that sex of rearing may be the most important determinant of a person's gender identity.

At present, however, the clinical literature is deeply divided on the best way to manage cases of traumatic loss of the penis during infancy.^{2,18,34–37} On the one hand, there is resort to the orthodox policy advocated by Money and colleagues.^{1,5} On the other hand, Diamond and Sigmundson¹⁸ suggest that such patients be reared as boys, despite the absence of a penis, on the grounds that the central nervous system has been biased in a masculine direction, in part because of normal prenatal androgenization. They recommend that “[s]urgery to repair any genital problem, although difficult, should be conducted in keeping with this paradigm.”¹⁸ To some extent, the course of action in individual cases may be dictated partly by parental choice between these two competing models of psychosexual management. Additional study is clearly required to decide which model of management results in the best possible psychosocial and psychosexual adaptation for individuals so affected.

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