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GENDER IDENTITY DISORDER IN CHILDREN AND ADOLESCENTS

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■ **Abstract** Gender identity disorder entered the psychiatric nomenclature in the DSM-III in 1980. This article reviews three domains of empirical research on gender identity disorder in children and adolescents: diagnosis and assessment, associated psychopathology, and developmental trajectories.

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INTRODUCTION

This article provides a selective review of research on children and adolescents with sex-typed behavioral patterns that correspond to the diagnosis of Gender Identity Disorder (GID), as defined in the fourth edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM) (American Psychiatric Association 2000). Three domains are reviewed: diagnosis and assessment, associated psychopathology, and developmental trajectories. I refer the reader elsewhere for summaries of etiological research (Cohen-Kettenis & Pfäfflin 2003, Green 1987,

Zucker & Bradley 1995) and the clinical literature on therapeutics (Cohen-Kettenis & Pfäfflin 2003, Meyer-Bahlburg 2002, Zucker 2001a).

Some readers of this volume are probably aware that GID is currently very popular in the media. On television, episodes of *Chicago Hope*, *Cold Case*, *ER*, *Law & Order*, and *Party of Five* have all had story lines on the topic. Two critically acclaimed films, *Ma Vie en Rose* (My Life in Pink) in 1997 and *Boys Don't Cry* in 1999, have also focused an artistic gaze on the subject. Indeed, in the latter film, actress Hilary Swank won an Academy Award for her role as Brandon Teena. Teena (born Teena Brandon), a female-to-male transsexual from Nebraska, was raped and subsequently murdered in 1993 at the age of 21 after two of his male friends discovered that he was a biological female (Sloop 2000, Willox 2003). The print media has also given attention to GID, including articles in *Time* (Cloud 2000) and *Saturday Night* (Bauer 2002). And, most recently, the *Oprah Winfrey Show* (12 May 2004) featured several prepubertal "transgendered" children and their families, thus providing exposure of their situation to over 20 million viewers in the United States alone.

A BRIEF OVERVIEW

The study of gender identity disorders in children and adolescents began 45 years ago, with a seminal paper by Green & Money (1960). Over this period, the field has attracted only a small number of researchers, perhaps reflecting the relatively low prevalence of GID when compared to other conditions that affect children, such as attention-deficit/hyperactivity disorder or the anxiety disorders. The number of clinicians with extensive experience in the field is also rather modest. At the time of this writing, there are, to my knowledge, only three specialized gender identity clinics worldwide for children and adolescents: one in Amsterdam (recently relocated from Utrecht), one in London, and my own clinic in Toronto. In contrast, there have been many more specialized gender identity clinics for adults, although the number of university or hospital-based programs has dwindled, particularly in North America (Petersen & Dickey 1995). Research on gender identity disorders, however, has profited from advances in allied areas, including studies of normative gender development, sexual orientation, and physical intersex conditions. When one takes into account this additional research on psychosexual differentiation, the literature is reasonable in size to keep a specialist sufficiently busy.

As a term, gender identity was introduced into the professional lexicon by Hooker and Stoller almost simultaneously in the early 1960s (Money 1985). Stoller (1964), for example, coined the term "core gender identity" to describe a young child's developing "fundamental sense of belonging to one sex" (p. 453). This term was adopted by cognitive-developmental psychologists, such as Kohlberg (1966), who defined gender identity as the child's ability to accurately discriminate males from females and then to identify his or her own gender status correctly—a task considered to be the first stage in "gender constancy" development. The

end state is the knowledge of gender invariance (for review, see Martin et al. 2002).

A child's acquisition of gender identity is, however, more than a cognitive milestone—it is also surrounded by affective significance. Many youngsters take the knowledge that they are boys or girls quite seriously; some researchers have shown that young children display evidence of an "in-group" bias, attributing more favorable attributes to their own sex than to the other (Ruble & Martin 1998). As noted by Fagot & Leinbach (1989), however, the role of affect in early gender identity formation has been relatively neglected by developmentalists. For clinicians, however, affect has always been a cornerstone in their thinking about early gender identity formation. For example, the term "gender dysphoria" was used with increasing regularity to describe patients, particularly adults, who felt so unhappy about their biological status as males or females that they sought both hormonal and surgical sex-reassignment in order to align the body with their psychological gender (Fisk 1973). Once research-clinicians began to look, it became apparent that children could also experience profound unhappiness about their status as boys or as girls (Green 1974, Stoller 1968).

DIAGNOSIS AND ASSESSMENT

In contemporary psychiatric nosology, GID diagnoses appeared for the first time in the DSM-III (American Psychiatric Association 1980) and contained two diagnoses [Gender Identity Disorder of Childhood (GIDC) and Transsexualism], which were to be used for children and adolescents/adults, respectively. In DSM-III-R (American Psychiatric Association 1987), a second diagnosis was added for use with adolescents and adults: Gender Identity Disorder of Adolescence or Adulthood, Nontranssexual Type (GIDAANT). For DSM-IV (American Psychiatric Association 1994), the Subcommittee on Gender Identity Disorders (Bradley et al. 1991) made two substantive recommendations. First, the GIDAANT diagnosis was "sunsetted," since its validity as a subtype of GID was arguable; second, the DSM-III and DSM-III-R diagnoses of GIDC and Transsexualism were collapsed into one overarching diagnosis, Gender Identity Disorder, with separate criteria sets for children versus adolescents and adults that reflected age-related, developmental differences in clinical presentation. Table 1 shows the diagnostic criteria for GID as they appear in DSM-IV.

Reliability and Validity

The clinical research literature has paid very little attention to reliability of diagnosis for GID. One study showed that, for children, clinicians can reliably make the diagnosis (Zucker et al. 1984), but, to my knowledge, no study has evaluated the reliability of the diagnosis for adolescents.

For adolescents, this may reflect the general dearth of empirical research when compared to their child counterparts with GID (Cohen-Kettenis & Pfäfflin 2003).

TABLE 1 Diagnostic and Statistical Manual-IV diagnostic criteria for gender identity disorder[a]

A. A strong and persistent cross-gender identification (not merely a desire for any perceived cultural advantages of being the other sex)
   In children, the disturbance is manifested by at least four (or more) of the following:
   1. Repeatedly stated desire to be, or insistence that he or she is, the other sex
   2. In boys, preference for cross-dressing or simulating female attire; in girls, insistence on wearing only stereotypical masculine clothing
   3. Strong and persistent preferences for cross-sex roles in make-believe play or persistent fantasies of being the other sex
   4. Intense desire to participate in the stereotypical games and pastimes of the other sex
   5. Strong preference for playmates of the other sex
   In adolescents and adults, the disturbance is manifested by symptoms such as a stated desire to be the other sex, frequent passing as the other sex, desire to live or be treated as the other sex, or the conviction that he or she has the typical feelings and reactions of the other sex

B. Persistent discomfort with his or her sex or sense of inappropriateness in the gender role of that sex
   In children, the disturbance is manifested by any of the following: in boys, assertion that his penis or testes are disgusting or will disappear or assertion that it would be better not to have a penis, or aversion toward rough-and-tumble play and rejection of male stereotypical toys, games, and activities; in girls, rejection of urinating in a sitting position, assertion that she has or will grow a penis, or assertion that she does not want to grow breasts or menstruate, or marked aversion toward normative feminine clothing
   In adolescents and adults, the disturbance is manifested by symptoms of such as preoccupation with getting rid of primary and secondary sex characteristics (e.g., request for hormones, surgery, or other procedures to physically alter sexual characteristics to simulate the other sex) or belief that he or she was born the wrong sex

C. The disturbance is not concurrent with a physical intersex condition

D. The disturbance causes clinically significant distress or impairment in social, occupational, or other important areas of functioning

[a]Reprinted with permission of American Psychiatric Association.

If one considers the diagnostic criteria as they pertain to adolescents and adults, it can be seen in Table 1 that the Point A criteria contain four possible indicators of a "strong and persistent cross-gender identification": a stated desire to be the other sex; frequent passing as the other sex; desire to live or be treated as the other sex; and the conviction that he or she has the typical feelings and reactions of the other sex. The Point B criteria contain two possible indicators of a "persistent discomfort with [one's sex] or sense of inappropriateness in the gender role of that sex": preoccupation with getting rid of primary and secondary sex characteristics or the belief that one was born the wrong sex.

From a clinical practice perspective, one could argue that the application of the diagnostic criteria is not particularly difficult, since it is relatively uncommon, at least in specialized child and adolescent gender identity clinics, to encounter an

adolescent who has only very mild gender dysphoria. However, this may well not be the case in general clinical practice. Thus, it is important to keep in mind that the indicators of GID are meant to capture a "strong and persistent cross-gender identification" (Point A) and a "persistent discomfort" with one's gender (Point B), not transient feelings. Unfortunately, the DSM criteria are somewhat vague in assisting the clinician in making this distinction. Consider two examples from the Point A criteria. Note, for example, that the first indicator of the Point A criterion refers to a "stated desire" to be the other sex and that the third indicator refers to a "desire to live or be treated as the other sex." It is unlikely that these indicators were intended to reflect an episodic desire to be of the other sex or to live as the other sex but, for some reason, neither indicator included a specific reference to persistence or intensity. On this point, revisions to the criteria appear warranted.

From a practical perspective, however, it could be argued that the odds of making a misdiagnosis of GID are probably not that high, primarily because normative data suggest that the frequent wish to be of the opposite sex in both referred and nonreferred samples is quite rare. For example, in the standardization of the Child Behavior Checklist (CBCL), a commonly used parent-report measure of behavior problems in children and adolescents, the percentage of adolescent boys and girls (ages 12–16) whose parents endorsed the item "Wishes to be opposite sex" was extremely low. In the original version of the CBCL, data were available for three age groupings (4–5 years, 6–11 years, and 12–16 years) divided by sex. For several of these age $\times$ sex groupings, the percentage of parents who endorsed this item was 0 (see Achenbach & Edelbrock 1983, p. 33). Thus, against base rates, even a periodic desire to become a member of the opposite sex is quite atypical.

The DSM-IV criterion with regard to the "preoccupation" with one's primary and secondary sex characteristics (Point B-2) reflects well the adolescent expression of gender dysphoria as it pertains to discomfort with somatic sex, since the distress over physical sex markers is so pervasive. Nonetheless, even here one has to exert some caution in making the diagnosis. For example, a study by Lee (2001) found a great deal of overlap in feelings of "anatomic dysphoria" among female-to-male transsexual women and self-identified "butch" lesbians. Similarly, caution in differential diagnosis is called for because of the recently described phenomenon of "tranny boys" among young lesbian women (see, e.g., McCarthy 2003), in which, for example, there appears to be a desire for "partial" sex reassignment (e.g., mastectomy, but not masculinizing hormone treatment or the reverse).

As noted above, there is only one clinician-based reliability study of GID in children; however, a much more extensive literature has examined its discriminant validity. Over the past 30+ years, a variety of measurement approaches have been developed to assess the sex-typed behavior in children referred clinically for GID, including observation of sex-typed behavior in free-play tasks, on semiprojective or projective tasks, and on a structured Gender Identity Interview schedule. In addition, several parent-report questionnaires pertaining to various aspects of sex-typed behavior have been developed. In this line of research, several comparison groups have typically been utilized: siblings of GID probands, clinical controls,

and nonreferred (or "normal") controls (for a summary and review of measures, see Zucker 1992, 2005).

The results of these studies have demonstrated strong evidence for the discriminant validity of the various measures. Two examples are provided. The first is the Gender Identity Interview for Children, which contains 11 items. Each item is coded on a three-point response scale. Based on factor analysis, Zucker et al. (1993) identified two factors, which were labeled Affective Gender Confusion (seven items) and Cognitive Gender Confusion (four items), and which accounted for 38.2% and 9.8% of the variance, respectively. An item example from the first factor is, "In your mind, do you ever think that you would like to be a girl (boy)?" and an item example from the second factor is "Are you a boy or a girl?" Both mean factor scores significantly differentiated gender-referred probands[1] (n = 85) from controls (n = 98). Cutoff scores of either three or four deviant responses yielded high specificity rates (88.8% and 93.9%, respectively), but lower sensitivity rates (54.1% and 65.8%, respectively).

The second is the Gender Identity Questionnaire for Children (GIQC), a parent-report questionnaire (Johnson et al. 2004). The GIQC consists of 16 items pertaining to various aspects of sex-typed behavior that are reflected in the GID diagnostic criteria, each rated on a five-point response scale. A factor analysis based on 325 gender-referred children and 504 controls (siblings, clinic referred, and nonreferred), with a mean age of 7.6 years, identified a one-factor solution containing 14 items, accounting for 43.7% of the variance. The gender-referred children had a total deviant score that was significantly more than that of the controls, with a large effect size of 3.70, using Cohen's *d*. With a specificity rate set at 95% for the controls, the sensitivity rate for the probands was 86.8%.

Threshold Versus Subthreshold Cases

The majority of clinic-referred samples of gender-referred children have been deemed to meet the complete DSM criteria for GID based on clinician diagnosis. For example, in a cross-clinic, cross-national study of gender-referred children (total N = 488) in Toronto and Utrecht, The Netherlands (Cohen-Kettenis et al. 2003), the percentage that met the complete DSM criteria for GID was 67.0%. Clinically, it has been noted that the majority of subthreshold cases likely met the complete criteria at a younger age, but not at the time of assessment (Zucker & Bradley 1995).[2] Some critics have, however, expressed concern that the DSM criteria may not adequately differentiate children with GID from those children

[1]By "gender-referred," I mean children who are referred clinically for concerns about their gender identity development.

[2]It is not unusual in clinical practice to encounter children who are subthreshold for a psychiatric diagnosis. Movement from threshold to subthreshold can be due to several factors, including variation in natural history and prior efforts to treat the condition (with or without formal professional help).

who merely show a pattern of extreme "gender nonconforming" behavior but who are not "truly" GID (e.g., Corbett 1996, Haldeman 2000, Jalas 2003, Richardson 1996).

Comparative analysis of threshold versus subthreshold cases is important for two reasons. First, using external measures, it can indicate whether the DSM criteria reliably distinguish between these two diagnostic subgroups; in other words, the central issue is one of identifying the boundary for psychiatric disorder (cf. Kendler 1999). Second, if there is evidence that a valid distinction can be made, one can evaluate whether or not the subgroups differ in other ways, such as variation in long-term developmental trajectories and putative etiological factors.

In one study, Zucker & Bradley (1995) reported that the children who met the complete DSM criteria for GID ($n = 113$) were significantly younger, of a higher social class background, and more likely to come from an intact, two-parent family than were the children who were subthreshold for GID ($n = 80$). The two subgroups did not differ significantly with regard to sex composition and IQ. To test which variables, if any, contributed to the correct classification of the subjects in the two diagnostic groups, a discriminant function analysis was performed. Age, sex, IQ, and marital status contributed to the discriminant function, with age showing the greatest power. In the threshold group, 82.6% were correctly classified, and in the subthreshold group, 68.8% were correctly classified.

Several data sets have examined whether or not the two diagnostic subgroups differed on various measures of sex-typed behavior. As summarized in Zucker & Bradley (1995), the threshold group showed significantly more cross-gender behavior or less same-gender behavior than the subthreshold group on 11 of 17 measures, even after controlling for the demographic variables that also differed between the two subgroups. Two recent studies have yielded similar findings. Johnson et al. (2004) found that the subthreshold group ($n = 109$) had a mean score on the GIQC that was intermediate between that of the threshold cases ($n = 216$) and the controls. There was, however, clear evidence that the subthreshold group was "gender nonconforming" in that the effect size between their mean score and that of the controls was substantial (Cohen's d ranged from 1.44 to 3.28 when blocked by age groups [e.g., 3–5 years, 5–6 years, etc.]). In a sample of gender-referred children from Utrecht, Cohen-Kettenis et al. (2004) also found that the threshold cases ($n = 114$) had a significantly more deviant score on the GIQC than did the subthreshold cases ($n = 42$).

Taken together, these data suggest that even within a population of gender-referred children, the DSM criteria, when used categorically (threshold versus subthreshold), significantly differentiate the behavior of the subgroups on external measures. At present, however, the precise manner in which the two subgroups differ remains unclear. For example, consider the five behavioral criteria for Point A in Table 1. Several critics have argued that Points A-2 to A-5 index gender role behaviors, and thus may not be direct indicators of gender dysphoria per se. Given that DSM-IV specifies that four of the five indicators are required to meet the Point A criterion, it has been noted that a child can meet this criterion

in the absence of a verbalized cross-sex wish (A-1). Thus, the concern is that the Point A criteria, as they are currently constituted, may inappropriately "scoop in" youngsters who show extreme cross-gender behavior but are not necessarily gender-dysphoric (Bartlett et al. 2000, Wilson et al. 2002).

The rationale for condensing gender identity and gender role indicators in the Point A criterion has been described in detail elsewhere (Zucker et al. 1998). In part, the argument was based on factor analysis of an existing data set that showed these indicators to form one common factor, which was interpreted as indexing "a profound and persistent cross-gender identification" (see Table 1). [As an aside, a similar factor structure was detected in Johnson et al.'s (2004) study, described above.] At the time, it was argued that there may be a small subset of gender-referred children with substantial cross-gender identification (and inferred gender dysphoria), yet who did not voice directly the desire to be of the opposite sex, perhaps because of social desirability considerations.

Zucker et al. (1998) tested this conjecture by reexamining symptom ratings from parent interview data for 54 children who did not meet the DSM-III criteria for GIDC. In this analysis, we assessed whether these children would meet the proposed DSM-IV Point A criterion for GID. Because these children did not repeatedly state the desire to be of the opposite sex, we coded as present or absent the indicators for A2–A5. If all four indicators were present, these children would then meet the proposed cutoff for the Point A criterion.

For the 54 children, the mean number of indicators rated as present was 2.36 (SD = 1.33; range, 0–4). Of the 54 children, 16 (29.6%) had all four indicators. This would thus represent only a modest increase in the number of gender-referred children who would meet the Point A criterion in the absence of a frequently stated desire to be of the other sex. It should be noted that at the time these analyses were carried out (in the early 1990s), the Point B criterion for GID had not been finalized (Table 1). Thus, it is possible that some of the youngsters who now met the Point A criterion would not have met the Point B criterion, thus resulting in an even smaller number of new threshold cases.

Gender Identity Disorder as a Disorder?

Over the years, some critics have gone beyond quibbling with the diagnostic indicators for GID and have contested the general legitimacy of GID as a disorder (e.g., Isay 1997). In some respects, this is not surprising, as various DSM sexological diagnoses have been subjected to critical scrutiny, such as pedophilia (e.g., Green 2002) and premenstrual dysphoric disorder (e.g., Chrisler & Caplan 2002). Of course, homosexuality is the prototype example of this discourse and, indeed, debate about homosexuality as a psychiatric disorder played a central role in demarcating the boundary definitions of mental disorder in the DSM-III (Bayer & Spitzer 1982, Spitzer 1981, Zucker & Spitzer 2005).

Critics of GID as a disorder have made several claims: (*a*) GID is really nothing more than normal variation, albeit extreme, in gender-related behavior; (*b*) children

with GID show little evidence of distress and/or impairment and, if they do, it is not inherent to the condition, but merely a reaction to social disapproval; and (*c*) because GID in children is strongly predictive of a homosexual sexual orientation in adulthood (see section on Developmental Trajectories), its inclusion in DSM-III was nothing more than a veiled backdoor maneuver to prevent later homosexuality. As I have attempted to counter the last claim elsewhere (Zucker & Spitzer 2005), I will appraise here only the first two criticisms and suggest ways in which the diagnostic criteria for GID might be reformed in DSM-V.

GENDER IDENTITY DISORDER AS NORMAL VARIATION IN GENDER-RELATED BEHAVIOR The definition of what exactly constitutes a mental disorder vis-à-vis the DSM has had a long and complex history (Widiger & Clark 2000). Although the DSM-III borrowed heavily from Spitzer & Endicott's (1978) explication of mental disorder, it was conceded that "there is no satisfactory definition that specifies precise boundaries for the concept of 'mental disorder"' (American Psychiatric Association 1980, p. 5). As a contemporary example of this broader debate, one might note the controversy regarding the Axis II personality disorders, in which there is considerable argument regarding the appropriate cut point between normal variation in personality traits and disorder (see, e.g., Livesley 2003, Widiger 2000). Regarding GID, Meyer-Bahlburg (1985) identified the problem as demarcating the "zone of transition between clinically significant cross-gender behavior and mere statistical deviations from the gender norm" (p. 682).

Because of the putative conflation of gender identity dysphoria and gender role behavior, particularly in the Point A criterion, one could argue that reform of the criteria is called for. One relatively simple reform would be to raise the threshold for the Point A criterion and require the presence of all five indicators. Thus, it would be necessary for the child to systematically verbalize the wish to be of the opposite sex in order for the Point A criterion to be met.

A more radical reform would be to relegate the indicators of extreme cross-gender role behavior (A2–A5) to the text description of GID, with an explanation that they may not be sufficient, on their own, to indicate the presence of gender dysphoria. The diagnostic criteria could then be modified by the inclusion of multiple indicators of gender dysphoria. For example, in addition to assessing the presence of verbal statements about wanting to be of the other sex, one could evaluate the presence of other markers, such as reporting of confusion about whether or not one is a boy or a girl, expressed unhappiness about being a boy (or a girl), that one feels more like a member of the opposite sex, dreaming about being of the opposite sex, and so on. To explore this, new psychometric studies will be required: (*a*) factor analysis can be used to establish the coherence of these putative indicators of gender dysphoria; (*b*) this would be followed by tests of discriminant validity; and then (*c*) an empirical determination of the appropriate cutpoint for high sensitivity and specificity. If such an enterprise proved successful, perhaps this would allay concerns that children with extreme gender

nonconformity, but who are not truly gender dysphoric, are being inappropriately diagnosed.[3]

DISTRESS AND IMPAIRMENT As noted by Zucker (1992), the DSM-III-R did not provide guidelines regarding the assessment of distress in the Point A criterion ("persistent and intense distress" about being a boy or a girl) or the ways in which it might be distinct from other operationalized components in the criteria (namely the "desire" to be of the other sex). In the DSM-IV this problem persists, except that it is now located in Point D, and there is the additional problem of defining impairment.

The inclusion of a distress/impairment criterion ("a clinical significance criterion") (American Psychiatric Association 2000, p. 8) is not unique to GID; in fact, this criterion now appears in most of the DSM-IV diagnoses. Very little empirical work preceded the introduction of the criterion (Spitzer & Wakefield 1999). Indeed, the DSM-IV states that the assessment of distress and impairment is "an inherently difficult clinical judgment" (American Psychiatric Association 2000, p. 8). Some critics have argued that the distress and/or impairment that clinicians observe in people with GID are not inherent to the condition, but merely a response to the reaction of others. Because of this, it is claimed that GID does not meet the DSM's own definition of a mental disorder (Bartlett et al. 2000).

Some theorists might agree with this critique. Stoller (1968), for example, argued that GID in boys was ego-syntonic and that they became distressed only when their cross-gender fantasies and behaviors were interfered with. Stoller's conceptualization was, however, a psychodynamic formulation: He believed that GID was ego-syntonic because the familial conditions that produced it were systemically syntonic. Thus, it is likely that Stoller would have argued that the DSM's conceptualization of distress was, and is, too narrow. Others have argued that cultural acceptance of extreme cross-gender feelings results in the absence of any distress (Newman 2002) and, at least in adults, the transition from one gender to the other proceeds with remarkably little complication (Bartlett & Vasey 2004).

Other theorists, however, have argued otherwise. For example, Coates & Person (1985) claimed that GID was a "solution" to specific forms of psychopathology in the child, particularly separation anxiety and "annihilation" anxiety, that were induced by familial psychopathology. It is conceivable that both the "primary" and "secondary" views are correct or that one or the other better fits individual cases.

One difficulty that remains is how best to operationalize the concept of distress, but this holds true not only for GID but also for all of the other childhood

[3]As an aside, I point out here that the critics have never actually provided empirical demonstration of inaccurate diagnosis. Haldeman (2000), for example, claimed that "it is conceivable that a child could be diagnosed with GID exclusively on the basis of preference for gender atypical activities or play objects," (p. 195) or that "any boy who, for example, displays an even passing interest in art, music or cooking could, conceivably, be diagnosed as GID" (p. 198). Neither of these assertions is true (Zucker 2001b).

psychiatric conditions that include the distress/impairment criterion. A simple example of this is Separation Anxiety Disorder (SAD). If a 5-year-old with SAD shows chronic distress at having to attend kindergarten and hence be away from his or her attachment figure, is the distress simply secondary to the socially imposed requirement of attending school or is it inherent to the condition? The distinction between socially induced and "in-the-person" distress may simply be a false dichotomy.

It is difficult to argue that cross-gender feelings and behaviors simply constitute normative variation or do not constitute an example of impairment if one considers the developmental adolescent or adult "end state" of GID, i.e., the strong desire to align the body via contrasex hormones and sex-reassignment surgery (in females: mastectomy, phalloplasty; in males: penectomy/castration, vaginoplasty) to the felt psychological state. The required physical interventions are simply too radical to be thought about otherwise. It seems to me that even in cultures where there is apparently considerable tolerance for "transgendered" individuals, there still must be in-the-person distress regarding the disjunction between somatic sex and felt psychological gender (Poasa et al. 2004). Otherwise, why would an individual choose to live in a cross-gendered role?

Consider some analogous phenomena. Some individuals express the strong desire to have a healthy limb amputated, in part because it is experienced as ego-alien and interferes with their "true" identity, which some have labeled Body Integrity Identity Disorder. Such individuals report feeling "incomplete" until one of their limbs is removed (First 2005). Along similar lines, some men desire castration and/or penectomy because the genitals are experienced as ego-alien and interfere with their "true" identity as a eunuch (Wassersug et al. 2004). Again, it is hard to imagine that the distress caused by these body image difficulties are simply variations in normative body image, along the lines that, say, wearing one's hair in a particular style results in a greater satisfaction than wearing it in another way. In these conditions, the distress and impairment (the disjunction between felt gender and somatic sex; the disjunction between one's sense of identity as an amputee and having four limbs; the disjunction between one's sense of identity as a eunuch and having male genitalia) appear rather profound and would all certainly qualify for the distress and/or impairment criteria that currently constitute the DSM definition of mental disorder. Of course, other kinds of dissatisfactions in body image exist, such as the desire for cosmetic surgery or achieving an ideal weight (see, e.g., Davis 1995, Ricciardelli & McCabe 2004, Thompson et al. 1999), in which the line between disorder and mere discontent is, perhaps, more fuzzy.

In children with GID, the assessment of body image disturbance, including sex-specific anatomic dysphoria (e.g., the desire for penectomy and castration in boys; the desire for mastectomy and phalloplasty in girls), has been much less well studied, perhaps because these parameters are less salient in prepubertal youngsters and/or because the topics are more sensitive to explore clinically with minors. In any case, further research on these parameters is needed.

There are several domains in which impairment might be manifest in children with GID. For example, children with GID seem to have more trouble with basic cognitive concepts concerning their gender than do other children. Zucker et al. (1993) found that children with GID were more likely than were controls to misclassify their own gender, and Zucker et al. (1999) provided additional evidence that children with GID appeared to have a "developmental lag" in the acquisition of gender constancy. Given the ubiquity of gender as a social category, this may well lead to affective confusion in self-representation and in social interactions. And, as noted in the next section, evidence indicates that children with GID have poorer peer relations than do controls and more general behavioral problems, which are possible indices of impairment.

SUMMARY When one evaluates adolescents and adults with GID, it is difficult to argue that their gender dysphoria (distress) is simply due to the reaction of others. It is the marked disjunction between somatic sex and psychological gender that causes their distress and motivates such individuals to seek out treatment. With children, though, the measurement of distress is more complicated; however, one can argue that these children manifest distress by virtue of their strong desire to become a member of the opposite sex (as expressed verbally or by their chronic enactment of cross-gender fantasies). Indeed, I would argue that the child's "fantasy solution" belief that he or she should become a member of the other sex is, ipso facto, a valid marker of distress. Unless one wants to argue that the desire to change sex is simply a variation of normal gender development, the symptoms of GID appear to be compatible with DSM ideas about distress and impairment.

ASSOCIATED PSYCHOPATHOLOGY

Comorbidity—the presence of two or more psychiatric disorders—occurs frequently among children referred for clinical evaluation. If the putative comorbid conditions actually represent distinct disorders, it is important to know, for various reasons, whether one condition increases the risk for the other condition or if the conditions are caused by distinct or overlapping factors (Caron & Rutter 1991).

Regarding GID, an interesting example of comorbidity comes from several case reports describing its co-occurrence with Pervasive Developmental Disorders (PDDs) (e.g., Mukaddes 2002, Perera et al. 2003, Williams et al. 1996). These case reports mesh with my own clinical experience, having systematically evaluated seven boys with comorbid GID and PDD over the past several years. One 6-year-old boy, for example, who had many of the classic features of PDD, including intense behavioral rigidity and obsessional preoccupations (e.g., with vacuum cleaners), had been insisting that he was a girl for the past three years and would introduce himself to other children using a girl's name. He would have catastrophic temper tantrums if reminded that he was really a boy. It is, of course, highly unlikely that GID causes PDD or the other way round. Rather, it is conceivable that the relation

between GID and PDD is linked by traits of behavioral rigidity and obsessionality. Indeed, many parents of children with GID report that their youngsters are obsessed by issues surrounding gender (Coates 1985).

The most systematic information on general behavior problems in children with GID comes from parent-report data using the CBCL (for other studies, see Zucker & Bradley 1995, pp. 103–106). On the CBCL, clinic-referred boys and girls with GID show, on average, significantly more general behavior problems than do their siblings and nonreferred children (Zucker & Bradley 1995; see also Cohen-Kettenis et al. 2003).

Given that the siblings and nonreferred children do not engage, on average, in marked cross-gender role behavior, one might construe these findings as evidence for a relation between cross-gender role behavior and general behavior problems. The situation, however, is clearly more complicated because demographically matched clinical controls (who, on average, show typical gender role behavior) show levels of behavior problems comparable to the children with GID (Zucker & Bradley 1995). Recent normative studies, however, indicate that both concurrent and predictive relations exist between measures of gender identity and psychological adjustment. In one study (Carver et al. 2003), children (mean age 11.5 years) were assessed with regard to their self-rated sense of "gender typicality" and "gender contentedness." In both boys and girls, lower levels of gender typicality and gender contentedness were significantly associated with more internalizing behavior problems and lower ratings of global self-worth and self-perceived social competence. Subsequently, Yunger et al. (2004) showed that low gender typicality and gender contentedness both predicted a decrease in psychological well-being at a one-year follow-up.

Patterns and Correlates of Behavior Problems

GENDER EFFECTS On the CBCL, boys with GID have a predominance of internalizing, as opposed to externalizing, behavioral difficulties whereas girls with GID do not (Cohen-Kettenis et al. 2003, Zucker & Bradley 1995). Edelbrock & Achenbach (1980) used cluster analysis to develop a taxonomy of profile patterns from CBCL data. Intraclass correlations were calculated and then subjected to centroid cluster analysis, from which profile types were identified and labeled. Intraclass correlations can range from –1.00 to 1.00 and a score of 0.00 represents the mean of the referred sample in the standardization study. Using this system, we found evidence for a clear internalizing pattern for boys with GID (Zucker & Bradley 1995, pp. 86–87). For 4- to 5-year-old boys, the mean intraclass correlation for Depressed-Social Withdrawal was 0.04. For 6- to 11-year-old boys, the mean intraclass correlations for Schizoid-Social Withdrawal and Schizoid were 0.04 and 0.16, respectively. For both age groups, there was considerable "distance" from externalizing profile types; for example, for the 4- to 5-year-old boys, the mean intraclass correlation for Aggressive-Delinquent was –0.42 and for the 6- to 11-year-old boys, the mean intraclass correlation for Hyperactive was –0.33. Two other studies have

demonstrated that boys with GID show high rates of separation anxiety traits (Coates & Person 1985, Zucker et al. 1996a).

AGE EFFECTS Zucker & Bradley (1995) found that increasing age was significantly associated with degree of behavior problems in boys with GID (depending on the metric, r's ranged from 0.28 to 0.42, all p's <0.001). This finding was replicated in the cross-national, cross-clinic comparative study by Cohen-Kettenis et al. (2003). In the Toronto sample, for example, only 26.1% of 3- to 5-year-old children had a CBCL sum score that was in the clinical range (>90th percentile) compared to 62.1% of 6- to 12-year-old children. The corresponding percentages for the Utrecht sample were 43.8% and 61.7%, respectively.

Zucker et al. (2002) provided further evidence for age effects when comparing their samples of GID children and adolescents. The latter had significantly more behavior problems, even when controlling for demographic differences (e.g., IQ, social class) between the two groups. Using Cohen's d, effect sizes for the CBCL metrics ranged from 0.55 to 0.93. In the adolescent group, 84.7% had a CBCL sum score that was in the clinical range.

One explanation for these age effects pertains to the role of peer ostracism. It has been noted for some time that children with GID experience significant difficulties within the peer group. Based on maternal ratings, Green (1976) found that 18% of the boys were "rejected" by the peer group and 38% were "voluntary loners," and the rejected boys were significantly older than the loners and the remaining boys who were not judged to have peer relationship problems (reported in Zucker et al. 1995). Given evidence from normative studies for the increasing salience, with age, of sex-typed behavior in children's friendships (see Zucker et al. 1995), the role of peer social ostracism may be a factor that mediates the age effect for behavior problems.

Using a composite index of poor peer relations derived from three CBCL items (alpha = 0.81), Zucker et al. (1997) showed that children with GID had significantly more peer relationship difficulties than did their siblings, even when controlling for overall number of behavior problems. Cohen-Kettenis et al. (2003) found that in both Toronto and The Netherlands, boys with GID had significantly poorer peer relations than did girls with GID, consistent with normative studies that show that cross-gender behavior in boys is subject to more negative social pressure than is cross-gender behavior in girls (Zucker et al. 1995). Nonetheless, Cohen-Kettenis et al. (2003) found that poor peer relations was the strongest predictor of CBCL behavior problems in *both* GID boys and girls, accounting for 32% and 24% of the variance, respectively. This finding suggests that social ostracism within the peer group may well be a potential mediator between cross-gender behavior and behavior problems.

An observational study by Fridell (2001) provided more direct support for the idea that the cross-sex-typed behavior of boys with GID may influence how well they are liked by other children. Fridell created 15 age-matched experimental play groups consisting of one boy with GID and two nonreferred boys and two

nonreferred girls (age range 3–8 years). During these sessions, the children in the groups had the opportunity to play with masculine, feminine, and gender-neutral toys. After two 60-minute play sessions, conducted a week apart, each child was asked to select his or her favorite playmate from the group. The nonreferred boys chose most often the other nonreferred boy as their favorite playmate, thus indicating a distinct preference over the boy with GID. The nonreferred girls chose the other girl as their favorite playmate, thus showing a relative disinterest in either the boy with GID or the two nonreferred boys.

SUMMARY In summary, children with GID show, on average, as many other behavioral problems as do clinic-referred children. Although these general behavior problems may contribute to their difficulties in the peer group, like other children with behavior problems (Schneider 2000), it is likely the case that their marked cross-gender behavior is particularly salient in eliciting negative reactions from their peers.

Although we have shown that poor peer relations is likely an important correlate of general behavior problems in children with GID, this is not meant to imply that it is the only source of these difficulties. Other research, for example, has shown that CBCL behavior problems in GID boys are associated with a composite index of maternal psychopathology. This may reflect generic, nonspecific familial risk factors in producing behavior problems in general (Zucker & Bradley 1995), and the predominance of internalizing psychopathology may reflect familial risk for affective disorders (e.g., Marantz & Coates 1991) and temperamental features of the boys. Thus, it is likely the case that there are both specific and general risk factors involved in accounting for the general behavioral problems of children with GID.

DEVELOPMENTAL TRAJECTORIES

Adolescents and adults with GID, particularly those who have a homosexual sexual orientation (i.e., sexual attraction to members of one's birth sex), invariably recall a pattern of cross-sex-typed behavior during childhood that corresponds to the DSM criteria for GID (Green 1974, Zucker et al. 2004). Indeed, this pattern of recalled sex-typed behavior launched the first generation of prospective studies in the 1960s designed to identify behaviorally children who might be "at-risk" for the subsequent development of transsexualism (GID) in adulthood.

As these first-generation studies were being carried out, another line of research showed that adults with a homosexual sexual orientation, unselected for gender identity, were also more likely, on average, to recall patterns of childhood cross-sex-typed behavior when compared to their heterosexual counterparts. Bailey & Zucker (1995) analyzed by meta-analysis all retrospective studies that used some measure of childhood sex-typed behavior and that made a quantitative comparison between heterosexual and homosexual same-sex adults. A total of 41

studies were identified, which yielded 48 independent effect sizes: 32 compared heterosexual and homosexual men and 16 compared heterosexual and homosexual women. Using Cohen's *d*, there were, on average, substantial differences in patterns of recalled childhood sex-typed behavior between heterosexual and homosexual adults. Both homosexual men and women recalled more cross-sex-typed behavior during childhood than did their heterosexual counterparts (respective *d*'s were 1.31 and 0.96). Subsequent to the Bailey & Zucker meta-analysis, similar findings have been reported in 14 other studies (summarized in Zucker et al. 2004).

Based on these retrospective studies, then, one might predict that children with GID (who fall at the extreme end of a continuum of cross-gender identification) followed prospectively should be disproportionately likely to have either persistent GID (with a homosexual sexual orientation) or a homosexual sexual orientation (with GID desistance).

Follow-up Studies of Boys

To date, Green's (1987) study constitutes the most comprehensive long-term follow-up of behaviorally feminine boys, the majority of whom would likely have met DSM criteria for GID. His study contained 66 feminine and 56 control boys (unselected for gender identity) assessed initially at a mean age of 7.1 years (range 4–12). Forty-four feminine boys and 30 control boys were available for follow-up at a mean age of 18.9 years (range 14–24). The majority of the boys were not in therapy between assessment and follow-up. Sexual orientation in fantasy and behavior was assessed by means of a semistructured interview. Kinsey ratings were made on a seven-point continuum, ranging from exclusive heterosexuality (a Kinsey "0") to exclusive homosexuality (a Kinsey "6") (Kinsey et al. 1948). Depending on the measure (fantasy or behavior), 75% to 80% of the previously feminine boys were either bisexual or homosexual (Kinsey ratings between 2 and 6) at follow-up versus 0% to 4% of the control boys. Green also reported on the gender identity status of the 44 previously feminine boys. He found that only one youth, at the age of 18 years, was gender-dysphoric to the extent of considering sex-reassignment surgery.

Data from six other follow-up reports on 55 boys with GID were summarized by Zucker & Bradley (1995, pp. 285–286). At follow-up (range 13–26 years), 5 of these boys were classified as transsexual (with a homosexual sexual orientation), 21 were classified as homosexual, 1 was classified as a (heterosexual) transvestite, 15 were classified as heterosexual, and 13 could not be rated with regard to sexual orientation. If one excludes the 13 "uncertain" cases in these six studies, then 27 (64.2%) of the remaining 42 cases had "atypical" (i.e., homosexual, transsexual, or transvestitic) outcomes. In these studies, the percentage of boys who showed persistent GID was higher than that reported by Green (11.9% versus 2.2%, respectively), but the percentage who were homosexual (62.1%) was somewhat lower.

Zucker & Bradley (1995, pp. 290–297) reported preliminary follow-up data on a sample of 40 boys first seen in childhood (mean age at assessment 8.2 years; range 3–12). At follow-up, these boys were, on average, 16.5 years (range 14–23). Gender identity was assessed by means of a semistructured clinical interview and by questionnaire. Sexual orientation (for a 12-month period prior to the time of evaluation) was assessed for fantasy and behavior using the Kinsey et al. (1948) scale in a manner identical to Green's (1987) study.

Of the 40 boys, 8 (20%) were classified as gender-dysphoric at follow-up. Regarding sexual orientation in fantasy, 20 (50%) were classified as heterosexual, 17 (42.5%) were classified as bisexual/homosexual, and 3 (7.5%) were classified as asexual (i.e., they did not report any sexual fantasies). Regarding sexual orientation in behavior, 9 (22.5%) were classified as heterosexual, 11 (27.5%) were classified as bisexual/homosexual, and 20 (50%) were classified as asexual (i.e., they did not report any interpersonal sexual experiences).[4]

Cohen-Kettenis (2001) reported preliminary data on a sample of 56 boys first seen in childhood (mean age at assessment 9 years; range 6–12) and again later, when they had reached adolescence. Of these, 9 (16.1%) requested sex-reassignment, and all 9 had a homosexual sexual orientation (PT Cohen-Kettenis, personal communication, Feb. 1, 2003). Thus, the rate of GID persistence, at least into adolescence, was higher than that reported by Green (1987) and comparable to the rate obtained by Zucker & Bradley (1995), as noted above.

SUMMARY In taking stock of these outcome data, Green's (1987) study clearly shows that boys with GID were disproportionately, and substantially, more likely than were the control boys to differentiate a bisexual/homosexual sexual orientation. The other follow-up studies yielded somewhat lower estimates of a bisexual/homosexual sexual orientation. In this regard, at least one caveat is in order. In the Zucker & Bradley (1995) follow-up, the boys were somewhat younger than were the boys followed up by Green, so their lower rate of a bisexual/homosexual sexual orientation outcome should be interpreted cautiously, as one would expect these youth to underreport an atypical sexual orientation due to social desirability considerations (see Zucker 1988). But even these lower rates of a bisexual/homosexual sexual orientation are substantially higher than the currently accepted base rate of about 2% to 3% of a homosexual sexual orientation in men that has been identified in recent epidemiological studies (Laumann et al. 1994).

A more substantive difference between Green's (1987) study and the other follow-up reports pertains to the persistence of gender dysphoria. Both Zucker & Bradley (1995) and Cohen-Kettenis (2001), for example, found higher rates

[4]These data are revised from Zucker & Bradley (1995) based on additional information from four boys who had recontacted us at some point after their initial follow-up evaluation: All four of these boys reported an exclusive homosexual sexual orientation in both fantasy and behavior (representing a change from either an asexual or heterosexual sexual orientation) and one boy was gender-dysphoric and living as a woman (see Zucker 2004).

of persistence than did Green. At present, the explanations for this are unclear. One possibility pertains to sampling differences. Green's study was carried out in the context of an advertised research study whereas the Zucker & Bradley and Cohen-Kettenis samples were clinic-referred. Thus, it is conceivable that their samples may have included more extreme cases of childhood GID than in the sample ascertained by Green. Because the Amsterdam and Toronto groups now have substantially larger samples on which to complete outcome studies, it should be possible to carry out within-group analyses (cf. McNeil & Kaij 1979) to identify predictor variables (e.g., with regard to persistent GID versus desistent GID). Such information is urgently needed in order to understand the variation in gender identity and sexual orientation outcomes within a population of children referred for gender identity problems.

Follow-up Studies of Girls

Unfortunately, the long-term follow-up of girls with GID remains very patchy. In part, this reflects the lower rate of referred girls compared to referred boys with GID in child samples: 1:4.67 in one study (Cohen-Kettenis et al. 2003). Green (1979) reported on the adolescent or young adulthood outcome of four girls who were seen in childhood: one was transsexual, with a homosexual sexual orientation; the remaining three did not have apparent GID; of these, two reported a heterosexual sexual orientation in fantasy (but were not sexually active), and the other reported no sexual feelings. Cohen-Kettenis (2001) reported preliminary data on a sample of 18 girls first seen in childhood (mean age at assessment 9 years; range 6–12) and who had now reached adolescence. Of these, eight (44.4%) requested sex-reassignment, and all had a homosexual sexual orientation (PT Cohen-Kettenis, personal communication, Feb. 1, 2003). Thus, the rate of GID persistence, at least into adolescence, was high (and much higher than the rate of persistence for her GID boys).

In my own clinic, from a total sample of 60 girls, we have, to date, outcome data on eight girls, originally assessed at a mean age of 9 years (range 3–12) and followed-up at a mean age of 20 years (range 15–28) (for a description of the assessment procedure, see Zucker & Bradley 1995, pp. 290–292). Of these eight girls, three had persistent GID (at follow-up ages of 17, 17, and 24 years, respectively), two of whom had a homosexual sexual orientation and the third of whom was asexual. Of the remaining five girls, three had a homosexual sexual orientation and two had a heterosexual sexual orientation, and none had co-occurring gender dysphoria.

Although these data are preliminary, it appears that there is a range of outcomes. However, it is clear that the rates of GID and a homosexual sexual orientation without co-occurring gender dysphoria are likely to be higher than the base rates of these two aspects of psychosexual differentiation in an unselected population of women.

Disjunctions Between Retrospective and Prospective Data

A key challenge for developmental theories of psychosexual differentiation is to account for the disjunction between retrospective and prospective data with regard to GID persistence: It is clear that only a minority of children followed prospectively show a persistence of GID into adolescence and young adulthood.

In some respects, the situation is comparable to that which has been found for other child psychiatric disorders. For example, adults with antisocial personality disorder invariably will have had a childhood history of oppositional defiant disorder and conduct disorder, an example of retrospective continuity (Robins 1978). Yet, the vast majority of children with oppositional defiant disorder and the majority of children with conduct disorder followed prospectively will not be diagnosed with antisocial personality disorder in adulthood (see, e.g., Lahey et al. 2000, Zoccolillo et al. 1992).

Regarding children with GID, we need to understand why, for the majority, the disorder apparently remits by adolescence, if not earlier. One possible explanation concerns referral bias. Green (1974) argued that children with GID who are referred for clinical assessment (and then, in some cases, therapy) may come from families in which there is more concern than is the case for adolescents and adults, the majority of whom did not receive a clinical evaluation and treatment during childhood. Thus, a clinical evaluation and subsequent therapeutic intervention during childhood may alter the natural history of GID. Of course, this is only one account of the disjunction and there may well be additional factors that might distinguish those children who are more strongly at risk for the disorder's continuation from those who are not. One possibility is that the diagnostic criteria for GID, at least as they are currently formulated, simply are not sharp enough to distinguish children who are more likely to show a persistence in the disorder from those who are not.

An additional clue comes from consideration of the concepts of developmental malleability and plasticity. It is possible, for example, that gender identity shows relative malleability during childhood, with a gradual narrowing of plasticity as the gendered sense of self consolidates as one approaches adolescence. Some support for this idea comes from follow-up studies of adolescents with GID, who appear to show a much higher rate of GID as they are followed into young adulthood.

To date, the best data on long-term outcome on adolescents come from the Dutch group. Cohen-Kettenis & van Goozen (1997) reported that 22 (66.6%) of 33 adolescents went on to receive sex reassignment surgery (SRS). At initial assessment, the mean age of the 22 adolescents who received SRS was 17.5 years (range 15–20). Of the 11 who did not receive SRS, eight were not recommended for it because they were not diagnosed with transsexualism (presumably the DSM-IV diagnosis of GID); the three remaining patients were given a diagnosis of transsexualism, but the "real-life test" (i.e., living for a time as the opposite sex prior to the institution of contrasex hormonal treatment and surgery) was postponed because of severe concurrent psychopathology and/or adverse social circumstances. These

data suggest a very high rate of persistence of GID, which is eventually treated by SRS. It should be noted that the persistence rate could be even higher than 66% since Cohen-Kettenis & van Goozen (1997) did not provide follow-up information on the 11 patients who were not recommended to proceed with the real-life test or were unable to implement it.

In another study, Smith et al. (2001) reported that 20 (48.7%) of 41 other adolescent patients went on to receive SRS. At initial assessment, the mean age of the 20 adolescents who received SRS was 16.6 years (range 15–19). Of the 21 who did not receive SRS (mean age 17.3 years; range 13–20), the reasons were similar to that reported in the earlier study. Data from Smith et al. (2001) suggest that a substantial number of the patients who did not receive SRS were still gender-dysphoric at the time of a follow-up assessment that occurred, on average, 4.3 years later.

Data from our own clinic are relatively consistent with the findings from the Dutch group. Zucker & Bradley (1995) reported that 19 (43.2%) of 44 GID adolescents had been referred for assessment to the adult gender identity clinic in our hospital because of a continued wish for SRS. Of these cases, three patients had received both hormonal and surgical sex-reassignment; a number of the others had been placed on cross-sex hormonal treatment, but had not yet been approved for surgery. Many of the remaining adolescents continued with a chronic gender dysphoria, being unable or unwilling to comply with the real-life test requirements for SRS (Meyer et al. 2001).

Childhood Sex-Typed Behavior and Sexual Orientation: Explaining the Linkage

Because the linkage between GID in childhood and a later homosexual sexual orientation is so strong, understanding the connection is important from both a theoretical and a clinical perspective. The most prominent biological explanation is that both sex-typed behavior in childhood and sexual orientation in adolescence/adulthood are joined together by some common factor or set of factors.

The literature on genetic females with a physical intersex condition called congenital adrenal hyperplasia (CAH), an inherited, autosomal recessive family of disorders of steroid hormone synthesis, is illustrative. The most common form of CAH, about 90% of cases, is caused by deficient 21-hydroxylase activity in the adrenal cortex. Because there is no negative feedback from cortisol, there is an increase in adrenocorticotropic hormone secretion, which produces a massive elevation of both cortisol precursors and androgens. The overproduction of androgenic steroids during fetal development causes genital ambiguity or masculinization in genetic females and presumably affects sex-dimorphic neural structures (for review, see Hines 2004).

Because genetic females with CAH are exposed prenatally to sex-atypical levels of androgen, it has been hypothesized that this predisposes to postnatal behavioral masculinization.

A large body of research has confirmed this hypothesis: Compared with same-sex controls, girls with CAH have higher rates of both masculine gender role behavior in childhood (Hines 2004) and bisexuality/homosexuality in adulthood (see, e.g., Zucker et al. 1996b). Thus, some researchers have argued that the shift in the direction of male-typical behavior with regard to both gender role and sexual orientation are caused by the excessive exposure to androgen during prenatal development.

Psychosocial perspectives on the linkage have varied. For example, Green (1987) conjectured that, compared with control boys, a feminine boy's lack of close relationships with other boys and with his father might result in "male affect starvation." Thus, in adolescence and adulthood, homoerotic contact is used in some compensatory manner to achieve closeness with other males. This scenario is an example of accounting for a within-sex difference in a behavioral outcome (in this instance, sexual orientation)—it is not clear if male affect starvation during childhood would also account for a girl's later sexual attraction to males.

In Bem's (1996) developmental theory of sexual orientation, in fact, it is proposed that similar mechanisms are operative in the sexual object choice of feminine boys and feminine girls (and masculine boys and masculine girls). Thus, Bem's account is not so much a deficit model, as implied by the term "affect starvation," but rather a difference model. Bem proposed that variations in childhood temperament influence a child's preference for sex-typical or sex-atypical activities and peers:

> These preferences lead children to feel different from opposite-sex or same-sex peers—to perceive them as dissimilar, unfamiliar, and exotic. This, in turn, produces heightened nonspecific autonomic arousal that subsequently gets eroticized to that same class of dissimilar peers: Exotic becomes erotic. (Bem 1996, p. 320)

For feminine boys and feminine girls, males are exotic, whereas for masculine boys and masculine girls, females are exotic.

Bem's (1996) theory of sexual orientation represents a prototype in trying to unite typical and atypical development. There are, however, many unanswered questions and alternative interpretations raised by the theory. For example, Bem placed great emphasis on temperamental factors that affect a child's preference for sex-typical or sex-atypical activities and friendships—an emphasis that might be disputed by some developmentalists (Ruble & Martin 1998). Empirical evidence for the emergence of specific erotic feelings following "heightened nonspecific autonomic arousal" is scant, although it is quite likely that the relevant tests can be obtained through an analysis of emerging sexual interactions within the preadolescent peer group. Nonetheless, Bem's theory is intriguing in that it implies a greater potential for malleability in sexual orientation development than is apparent in some of the biological theories. For example, if a feminine boy becomes more masculine in the course of his childhood, does this imply that the likelihood of later homoeroticism decreases? Conversely, if a feminine girl becomes more

masculine in the course of her childhood, does this imply that the likelihood of later homoeroticism increases?

Unfortunately, there is not much information available to answer these questions. Green (1987) compared on a number of childhood variables the feminine boys who were subsequently classified as bisexual or homosexual with the feminine boys who were subsequently classified as heterosexual. Although some feminine behaviors distinguished the two subgroups, a composite extent of femininity score only approached conventional levels of significance and only for the rating of sexual orientation in fantasy, not behavior. The lack of a stronger correlation is somewhat surprising, since one might have expected an association between the degree of cross-gender identification and long-term outcome; however, Green (1987) did find that the continuation of certain feminine behaviors throughout childhood was associated with later homosexuality. Thus, it may be that the persistence of these feminine behaviors is more important than their extent during the early childhood years.

CONCLUSION

This article has reviewed three domains regarding GID in children and adolescents: diagnosis and assessment, associated psychopathology, and developmental trajectories. Since the GID diagnosis first appeared in the DSM-III in 1980, we have seen the accumulation of a reasonable body of empirical data in each domain, but there remain substantial gaps and areas for future exploration. I have identified areas in which the diagnostic criteria for GID require a fresh examination. Understanding the associated psychopathology in children with GID requires a broad perspective, incorporating both general and specific risk factors. And last, more developmental outcome data are required to delineate better the natural history of GID.

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