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Gender dysphoria and gender variance in children – diagnostic and therapeutic controversies

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Summary

The phenomenon of atypical gender identity in adolescents and adults, in diagnostic classifications called gender identity disorder – transsexualism (ICD-10) or gender dysphoria (DSM-5), arouses interest among research communities both in Poland and elsewhere. At the same time, much less attention is paid to the same phenomenon occurring in childhood, leaving a gap of potential significance to a wide range of specialists including psychiatrists, psychotherapists, pediatricians and social workers. The aim of this paper is to provide a synthesis and present the current state of knowledge about children with atypical gender identity and their families. Available data pertaining to etiology, psychopathology and development of individuals experiencing gender dysphoria in childhood are analyzed. Further, the aim is to highlight the controversies surrounding diagnostic and therapeutic processes, considering that the vast majority of gender-dysphoric (GD) children appear to no longer experience the symptoms in adolescence and adulthood. Therefore, the paper presents some important areas of care and stresses the need for great caution and individual approach when working with GD children and their families.

gender dysphoria in children, gender variance, gender identity in childhood, atypical gender identity development

The diagnostic category of gender identity disorder (GID) of childhood was first introduced[1] into the DSM-III classification (1980) [1] and was included together with transsexualism and atypical gender identity disorder in the gender identity disorder parent category. The most important characteristic of GID is the discrepancy between anatomical sex and gender identity defined as a strong sense of being either male or female [1]. From the beginning, this category has provoked controversies both among experts and members of LGBT (lesbian, gay, bisexual, transgender) rights organizations. The introduction of GID into the DSM diagnostic classification system was seen as a veiled attempt to restore the category of homosexuality, previously excluded from DSM-II, allegedly to legitimize "preventive treatment" of homosexuality [2,3]. Other allegations, which are maintained in modern literature, concern the validity of the GID category. In the course of preparing the fifth DSM revision, some LGBT activists claimed that it is stigmatizing to label behaviors incongruent with gender stereotypes as disorder symptoms

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[1] The first diagnostic category to describe strong atypical gender identity in children was incorporated in ICD-9. The category "Disorders of psychosexual identity" was included in the chapter called "Sexual deviations and disorders" and defined as behavior displayed in psychosexually immature pre-pubertal individuals, similar to "transvestism" and "transsexualism" (ICD-9) [11].

and that it may cause suffering in individuals who display such behaviors [4].

Criticism also came from research communities. Barlett et al. [5] claim that GID does not meet contemporary mental disorder criteria. In their opinion, a serious limitation of studies on GID is that they fail to separate children whose distress is caused by cross-gender behaviors and those whose distress is directly related to biological sex characteristics (gender dysphoria). Ehrbar et al. [6] point to difficulties with distinguishing between these groups at diagnosis. According to Barlett et al. [5], only the latter can meet some criteria for mental disorder as specified by the American Psychiatric Association (APA).

Eventually, despite the opposition, GID in children was included in the category of sexual disorders in DSM-5 [7]. The name of the category was changed, though, to gender dysphoria (GD) and the emphasis was shifted from gender identity itself to the related distress experienced by an individual which causes significant deterioration in his or her quality of life. As stressed by the APA [7], gender dysphoria refers to emotional distress over "(…) a marked incongruence between one's experienced/expressed gender and assigned gender." According to the definition, gender dysphoria is a subjective disorder of mood/affect experienced by people whose psychological gender is incongruent with their biological sex [8]. To be diagnosed with GD, a child has to manifest, for at least 6 months, at least six of eight signs (concerning attitudes towards anatomy, attire and play stereotypically associated with the assigned gender) included in DSM-5 [7]. One of these signs must be criterion A1: "A strong desire to be of the other gender or an insistence that one is the other gender (or some alternative gender different from one's assigned gender)." The condition must also cause clinically significant distress and impairment of functioning in important areas of life (criterion B). GD can be diagnosed with or without co-occurring disorders of sexual development.

Controversies over gender dysphoria are ongoing at a time when the eleventh revision of ICD is being prepared for publication in 2018. A document issued by the Global Action for Trans Equality (GATE) [9] questions the validity of the GD category pertaining to children and objects to pathologizing manifestations of gender nonconformity. GATE also argues that therapeutic and medical interventions in children who diverge from gender stereotypes contradict the attitudes of respect towards and acceptance of sexual diversity declared by the World Health Organization. GATE proposes that healthcare professionals should use non-pathologizing categories listed in ICD-10 under "Factors influencing health status and contact with health services" (Z codes) when working with individuals displaying GD symptoms [10]. Drescher et al. [11] warn that grouping GD with ICD-10 Z codes, which are intended for normative phenomena, could make insurance providers and healthcare systems refuse to financially support GD individuals. Access to treatment requires diagnosis, which recognizes a given phenomenon (directly or indirectly) as pathological. In their opinion, it is highly unlikely that social ostracism towards children with GD is related solely to the medical diagnosis. Drescher et al. found that the community agrees that GD children should have access to specialist help, while the exclusion of GD category is problematic and counterproductive. They suggest that GD could be treated the same way as other natural phenomena (such as menopausal and female climacteric states (N95.1) or single spontaneous delivery (O80)), subject to medical treatment and included in classification systems to ensure patients' access to healthcare despite the fact that they are not pathological in any way.

Importantly, the term used in the source literature and by advocates of excluding gender dysphoria from diagnostic classification is a somewhat broader gender variance (GV), which is considered less pathologizing than either gender identity disorder or gender dysphoria [11]. In this paper we use gender dysphoria unless the historical perspective or original terminology require otherwise.

ETIOLOGY, PREVALENCE AND COMORBID DISORDERS

The process of gender identity development and gender dysphoria as well as factors that affect them remain largely unknown [12]. The impact of genetic and environmental factors is estimat-

ed based on studies conducted on twins. However, findings are ambiguous. Beijsterveldt et al. [12] indicate that intensification of cross-gender behaviors in children is a highly heritable characteristic – additive genetic factors accounted for about 70% of variance in their study. Similar conclusions were drawn by Coolidge et al. [13]. However, due to the low statistical power of the study, the hypothesis about the impact of environment on the development of cross-gender behaviors could not be discounted. A study by Knafo et al. [14] on a sample of 5799 pairs of twins aged 3 and 4 indicates that the impact of heritability and environment on the development of cross-gender behaviors was gender specific. In boys, environmental influences explained most of the variance of feminization (51–57%), whereas the impact of heritability was moderate (21–32%). In girls, the impact of heritability was significantly greater (42–50%), while the impact of environment was weaker (33–43%). Nevertheless, results varied depending on the definition adopted. In the group of girls with high masculinization and low feminization scores, the impact of heritability was high (65%), whereas environmental influence was of no consequence.

A biological explanation of the development of gender-related behavior is focused around the effect of sex hormones on prenatal development. Girls with congenital adrenal hyperplasia who were exposed to high androgen levels in their prenatal life displayed higher preference for toys targeted at boys and for the company of boys during playtime when aged 3 to 8. In boys with congenital adrenal hyperplasia no such effect was observed [15].

According to certain psychological theories, the desire to become a person of the opposite sex is explained by, for instance, compensatory response to trauma, disordered attachment to primary caregiver or separation disorder, where the child becomes symbolically symbiotic with the caregiver (mother or father). The desire to change one's gender is also considered a universal strategy in coping with developmental tasks that the child sees as beyond his or her coping capacity [16]. Some studies and theories postulate the existence of intense psychopathologies in mothers or both parents of GD children. However, both research and therapeutic communities remain far from agreement on this particular issue [16,17].

Gender identity cannot be formed under the sole influence of external factors (the so-called 're-education') even if such an attempt is initiated in the first year of life [18]. This suggests early biological determination of gender identity. Therefore, it should be assumed that gender identity develops as a result of biological and psychosocial factors interacting [16].

No epidemiological studies on GD children have been published so far. Thus, prevalence data had to be based on less sophisticated methods and have a certain margin of error [20]. In Dutch longitudinal studies conducted on a group of twins, behaviors atypical of a given sex were present in 3.2% boys and 5.2% girls aged 7 (n=14000). At the time of follow-up at the age of 10 (n=8500), these figures dropped to 2.4% and 3.3% [13]. However, neither the DSM nor the ICD criteria were applied to make the diagnosis. The prevalence of GD in children as far as the clinical diagnosis is concerned is estimated at below 1% [16,19]. Most studies report greater prevalence of cross-gender behaviors and GD in boys (up to 6 times as common as in girls) [7,19–21], but some indicate equal prevalence in both sexes [22].

Since the actual prevalence of gender dysphoria in the population is unknown, it remains unclear whether the disproportion between boys and girls reflects this prevalence or is rather related to other factors. Zucker & Lawrence hypothesize that this disparity might result from greater biological susceptibility in boys, as male sexual differentiation depends on androgen production in early stages of prenatal life [19].

Gender dysphoria is often related to behavioral issues and a high percentage of comorbid disorders. In studies conducted by Wallien et al. [23], 52% of children diagnosed with GD met criteria for other mental disorders: anxiety disorders were reported in 31%, behavioral disorders in 23% and mood disorders in 6%. In a study by Spack et al. involving a trial of 91 children and adolescents with GD referred to outpatient clinics, 44.3% had been diagnosed with other mental disorders, 37.1% received psychotropic medications and 21.6% manifested auto-aggressive behavior at the time of or prior to the study [22].

DEVELOPMENT TRAJECTORIES IN CHILDREN WITH GENDER DYSPHORIA

Investigating the developmental paths of individuals who experienced gender dysphoria in childhood appears to be crucial for accurate help and therapeutic interventions that would be in line with the rules of medical or psychological ethics. At the same time, due to the fact that not all caregivers of children with atypical gender identity can seek or choose to seek specialist help, one should bear in mind that data at our disposal reflect mostly clinical populations and refer to people who are patients of gender identity clinics.

Research studies from the Netherlands, Canada and the United States suggest that people with a history of gender dysphoria of childhood mostly do not experience symptoms in adolescence and adulthood [24–29]. Group-level factors that differentiate adolescent and adult transgender persisters from cisgender[2] desisters included higher prevalence of cross-gender behaviors [24,25], lower social status [25], older age of the child at the time of first diagnosis, and female biological sex [27,46]. Some researchers suggest that the plasticity of gender identity differentiation is greater in early childhood and decreases in adolescence, which might explain why childhood gender dysphoria does not manifest to the same extent in adolescence [27]. In the case of biological sex, it is difficult to assess whether developmental paths of boys and girls are indeed that different. However, as several times more boys than girls are referred to gender identity clinics, at present we have much more empirical data on the developmental trajectory in biological boys diagnosed with GID/GD at our disposal. This may suggest that the differences reported in this paper should be treated with caution, as they may result from the fact that study groups of biological girls and women who consented to participating in follow-up research were not representative, as well as from actual differences in the development of boys and girls diagnosed with GID/GD.

Of the biological girls aged 3–12 referred in the years 1975–2004 to a gender identity service in Toronto, Canada, 12% (N=3) of those reassessed at the age of 15–36 still demonstrated gender dysphoria [24]. In turn, a Dutch study [27] of girls with GID assessed at the age of 5–12 and reassessed at 16–25 noted persistent gender dysphoria in 50% (N=8) of subjects. As for psychosexual orientation determined based on fantasies, in a Canadian study 68.18% of teenage and adult non-dysphoric women exhibited heterosexual orientation exclusively, 27.27% manifested homo – or bisexual orientation, whereas 4.5% subjects were identified as asexual. Out of 3 subjects experiencing GD, 2 reported to be attracted to women and 1 was asexual [24]. In contrast, in a Dutch group all cisgender women were heterosexual, while all individuals experiencing GD were attracted to women [27]. As for biological boys diagnosed with GID/GD of childhood, recent studies noted that gender dysphoria persisted into adolescence and adulthood in 9–20% subjects [25,27,28]. Non-heterosexual orientation was more commonly observed among cisgender non-dysphoric men than among cisgender women, i.e. in 38-56% subjects [25,27,28]. As for biological boys and men with persistent GD, nearly all were attracted to men at follow-up [25,27]. Investigating the prevalence of specific developmental paths among 125 biological boys, Singh [25] indicated that in adolescence and adulthood cisgender-homo/bisexuality was most common (52.8% subjects), while over a third of assessed boys and men (33.6%) were cisgender and heterosexual. Persistent gender dysphoria combined with being sexually attracted to the same biological sex or both sexes was noted in 12.8% of subjects, with one of the dysphoric boys being attracted to the opposite biological sex. At the same time, only one of the studies recorded data on individuals who used some form of therapy or specialist consultations regarding gender identity in between individual diagnostic assessments [24].

TREATMENT APPROACHES

Although the APA Task Force on Treatment of Gender Identity Disorder [31] reached an agree-

[2] The term *cisgender* (in contrast to *transgender*) is used for people who identify with their assigned gender, i.e. their gender identity matches their biological sex [30].

ment on recommendations for therapeutic interventions in adults and adolescents with gender dysphoria already in 2012, numerous controversies and differing expert opinions hindered a similar consensus about therapy for prepubertal children at that time. Due to underlying assumptions and the scope of interventions applied, models of therapeutic work with GD/GV children and their families currently used vary significantly, ranging from corrective or normalizing approaches to affirmative or early transition models.

Interventions identified as corrective or normalizing [32] are based on a belief that therapeutic work in childhood can eventually eliminate dysphoric symptoms and help the child feel comfortable with his or her biological sex [17]. Incentives for building positive relations with peers and the same-sex parent, engaging in gender-typical or gender-neutral activities, as well as limiting cross-gender behaviors are accompanied by attempts aimed at identifying possible factors of disordered gender identity. At present, experts no longer acknowledge the "prevention" of non-heterosexual orientation as an ethical and valid justification of methods applied. Instead, they consider it reasonable to protect the child from social ostracism and complex medical procedures that they would be exposed to if gender dysphoria persisted [33]. Moreover, there is no empirical evidence to show that normalizing interventions can lead to an improvement in dysphoric conditions and the subjects' mental health [32]. Also, children who are not allowed to behave in a way congruent with their experienced gender identity may face intensified dysphoric feelings, comorbid symptoms and suicidal tendencies [34,35].

Contrary to the strategy applied at the clinic in Toronto [33], the Dutch approach [36] is not targeted directly at GD symptoms. Instead, therapeutic work focuses solely on the emotional, behavioral and family issues that may affect gender dysphoria in a child. Although children's cross-gender behavior is not restricted within the framework of the program, it is recommended that full social transition is deferred until adolescence. This recommendation constitutes a reasonable preventive measure in case a child wishes to return to a role congruent with his or her biological sex if dysphoria does not persist [37].

On the other end of the continuum, there are affirmative and early transition models that endorse the child's behavior and expression of preferred gender identity [32,35,38,39]. The affirmative approach assumes that a family system accepting and supporting the child in building his or her psychical resilience, as well as the child's immediate surrounding, constitute a space where the child can establish his or her true gender self in the course of further development. While early transition in children displaying a strong need for transition is advocated, at the same time parents are encouraged to remain open to and ready for the possibility that their child's development might take various directions including reidentification with his or her biological sex [39]. Provided no dysfunction within the family system or trauma history that could affect the child displaying GD symptoms is present, most radical supporters of childhood transition claim children should be allowed to function according to their innate (or what is considered innate) gender identity as early as possible [35]. This approach, which has been enthusiastically received by transgender adults, is a source of concern for experts who point out the relatively insignificant percentage of children with gender dysphoria/gender variance persisting into adolescence and adulthood, as well as a lack of studies on long-term consequences of early transition [40]. Regardless of the approach chosen, two factors are important: areas that play a significant role in working with this group of patients (based on clinical experience) and findings of studies into the needs of parents and transgender adults [41].

Table 1. Areas of work with gender dysphoric children and their families

Behavioral, emotional and relational difficulties related to gender dysphoria/variance
Psychoeducation in the area of the child's further psychosexual development
Supporting both the child and the family in coping with anxiety and insecurity regarding future development of the child's gender identity and sexual identity
Creating a safe space for discussing the child's feelings related to the preferred gender expression
Supporting parents in responding to the child's need of unconditional love regardless of the manifested gender expression and in building a common approach to the child's behavior
Supporting the family in deciding on revealing the child's GD/GV in specific contexts of functioning (distant relatives, friends, nursery, school, etc.)
Creating within the family a space for emotional processes – anxiety, denial, sadness, anger or disgust – experienced by parents in response to displays of atypical development of the child's gender identity and grieving related to the loss of an anticipated direction of the child's psychosexual development
Building up resilience and competence required for coping with bullying or other forms of discrimination of GD/GV children and their parents
Providing the family with knowledge on further medical and therapeutic possibilities in their home country and abroad
Assisting families in seeking out experts (pediatricians, psychiatrists, psychologists, endocrinologists, therapists, sexologists)

EARLY HORMONAL THERAPY

Both experts working in the field of atypical gender identity development and parents of GD children should be aware of possible future directions of the therapeutic process if gender dysphoria of childhood does not resolve until adolescence. In 2009, a Dutch research team [42] published recommendations for early hormonal therapy allowed in GD adolescents who satisfy the criteria of readiness for physiological change. The therapy involves gonadotropin-releasing hormone (GnRH) analogues aiming at inhibiting gonadotropin secretion and thus suppressing puberty. It is intended as a means of extending the time when it is possible to assess the direction in which dysphoria develops [16]. Early hormonal therapy is also one of the first steps undertaken towards sex reassignment [42]. Hembree et al. [42] recommend commencing the therapy upon the onset of pubertal physical changes, which ought to be confirmed by sex hormone levels (estradiol in girls and testosterone in boys), but no earlier than Tanner stage II or III (Tanner Scale is used for assessing the progression of pubertal changes). The physiological effects of therapy are completely reversible.

Benefits of suppressing puberty at an early stage include reduced psychological distress and lessened intensity of comorbid disorders, as well as improved social functioning [16,23]. Individuals who had taken GnRH analogues manifested a better cosmetic effect when further surgical intervention was applied [16,43]. However, the APA Task Force on Treatment of Gender Identity Disorder report stresses that long-term effects of early puberty-blocking therapy are unknown [31]. Though completely reversible in physiological terms, inhibited puberty can have a permanent effect on psychosexual development [16]. Suppressed sexual drive and lack of sexual experience typical of adolescence disturb the formation of sexual identity, orientation and preference, as well as self-assessment of functioning in interpersonal relations. The effect of this therapy on the persistence or disappearance of atypical gender identity remains unclear. Further, it was noted [9] that emotional and cognitive competencies vital for making a conscious decision to commence or reject the therapy are not sufficiently developed in adolescents. Not all specialists recommend early therapy with GnRH analogues [9]. Due to high percentage of desisters, the Royal College of Psychiatrists (UK) recommends that cases should be considered on an individual basis and that the appropriateness of initiating therapy at initial stages of adolescence should be assessed with caution [46].

SUMMARY

Despite many years of research and debate among specialists attempting to analyze the phenomenon of gender dysphoria in childhood, there are still no established standards of diagnostics and therapeutics. As childhood gender dysphoria, with its etiology, correlates and developmental trajectories, is so poorly understood, it is exceptionally difficult to decide on a proper course of action. Basing on studies conducted so far, it seems that gender dysphoria is more likely to persist into adolescence and adulthood in biological girls than in boys, whereas where it disappears, cisgender women more often prove to be heterosexual than cisgender men, approximately half of whom develop homo – or bisexual orientation. Nevertheless, one should bear in mind that patterns described above are observed at a group level, whereas no effective diagnostic method for predicting a future course of psychosexual development of an individual diagnosed with GID/GD of childhood has been developed as yet [45].

At present, an expert who undertakes work in the field of gender dysphoria in children cannot base their decision on the choice of a given work paradigm or findings of empirical studies dedicated to the effectiveness of specific therapeutic programs. As the topic is highly emotionally charged and making an objective judgment is exceptionally difficult, practitioners and researchers alike should carefully monitor both the available literature (which, admittedly, can be tainted by prejudice and dubious premises) and their own personal attitudes potentially influencing their work. This makes one reflect on the lack of overarching principles that would provide some sort of guidance in the process of supporting GD children and their families. It seems that these principles should allow for broadly understood safety of the child who experiences gender dysphoria and strengthening their psychological resilience. Experts who strive to act according to the child's best interest when providing their health care may wonder who is to decide what is best for the child – the child, the parents, the therapist or the society which imposes standards of appropriate gender expression. Is the child's best interest consistent with the best interest of a future adult? On the one hand, an in-depth diagnosis of the intensity of a child's dysphoric feelings and their possible reaction to having their expression congruent with experienced gender identity limited, as well as an assessment of environments in which the family and the child operate, may lead in certain cases to different conduct patterns developed together with the child's parents. On the other hand, strengthening the child's psychological resilience should constitute a universal component of interventions. A resilient child can better manage possible difficulties within the family with accepting his or her gender identity, better cope with diverse reactions of the environment in the face of an open expression of their gender dysphoria/gender variance, as well as conceal his or her gender identity in a context that may sometimes be threatening.

Further research is necessary to establish to what extent the previously observed prevalence of persistent and waning gender dysphoria was determined by natural developmental tendencies and to what extent it was affected by therapeutic interventions. Research must also investigate long-term outcomes of particular therapeutic approaches.

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