Chapter 1
General Introduction
Introduction

One of the first things we are curious about when we hear of a newborn child is the natal sex. We wonder “is it a boy or a girl?” With this information, we generally start imagining his or her future gender role; behaviors, attitudes, and personality traits that are typically attributed to, expected from, or preferred in a boy or a girl. In our culture, we expect, for example, that boys will have a greater preference for cars and show more rough- and tumble play than girls, whereas girls will have a greater preference for dolls and show more social interaction in play behavior than boys. At the same time, we rarely wonder what children’s future gender identity (their experience to be male, female or a different gender), will be like. A natal boy is expected to identify and label himself as a boy, and a girl is expected to identify and label herself as a girl. This is not surprising, since, for most children, gender identity and gender role expressions are largely congruent with each other and in line with their natal sex. However, not all children go through a normative or typical gender development. Some will not show gender role behaviors and/or not have a gender identity that is congruent with their natal sex.

Gender role and gender identity in childhood can be placed on the male-female continuum (see Figure 1). Depending on their positions on these continua, three groups can be distinguished. One group consists of the gender normative children. Their gender role and gender identity are largely in line with their natal sex. A second group consists of the gender variant children. These children may show gender role behaviors, interests and preferences, and may experience a gender identity which is congruent with their natal sex to a lesser extent than is the case in gender normative children. The expression of gender variance in these children is not necessarily associated with distress. This is in contrast to gender dysphoric children. These children show extreme and enduring forms of cross-gender behaviors, preferences and interests and may indicate that they want to be the other gender because they experience cross-gender identification. Some of these children may even express anatomic dysphoria (“I do not want to have a penis” or “I do not want to have breasts, when I grow up”). Children who have clinically significant distress or have a significant risk of distress, and/or may be impaired in important areas of functioning, thereby fulfilling the criteria for a DSM-IV-TR diagnosis of a Gender Identity Disorder (GID) (American

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1 Although it is conceivable that children have an identity outside the male-female continuum, thus far we did not come across children identifying as gender queer in our studies or in our clinical work.
Psychiatric Association 2000), often need clinical attention (Meyer-Bahlburg 2010).

Figure 1  Positions of gender normative, gender variant and gender dysphoric natal boys’ gender role and gender identity on the male-female continuum

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Gender normative boys Gender variant boys Gender dysphoric boys

GI - Gender identity, GR - Gender role

Epidemiology

Formal epidemiological studies on the prevalence (the presence or absence of a behavioral phenomenon in a representative sample of individuals) of gender variance and gender dysphoria in childhood do not yet exist. An estimate of the prevalence of gender variance can, however, be made on the basis of studies where the Child Behavior Checklist (CBCL) (Achenbach and Edelbrock 1983) was used. On the CBCL, a widely used parental report questionnaire that measures emotional and behavioral problems in children, two items are related to gender variance: Item 5 (“Behaves like opposite sex”) and item 110 (“Wishes to be of opposite sex”). Information from the Dutch normative sample of the CBCL shows that in children between the ages 4 to 11, both items are more frequently endorsed by parents of girls

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2 The CBCL version prior to 2001 consisted of the two items related to gender variance, the current version only consists of item 110.
than of boys; item 5; “Behaves like opposite sex” (either a 1 or a 2) in 2.6% of the boys and 5.0% of the girls, item 110; “Wishes to be of opposite sex” in 1.4% of the boys and 2.0% of the girls (Verhulst et al. 1996). These findings are in concordance with data from the normative sample of children between the ages 4 to 11 of the CBCL in North-America, in which item 5 was endorsed with a 1 for 3.8% and a 2 for 1.0% of the boys, and endorsed with a 1 for 8.3% and a 2 for 2.3% of the girls. Item 110 was endorsed with a 1 for 1.0% and a 2 for 0.0% of the boys, and endorsed with a 1 for 2.5% and a 2 for 1.0% of the girls (Achenbach and Edelbrock 1981; Zucker et al. 1997). Furthermore, a Dutch CBCL study on a sample of twins, who were 7 years of age, showed that item 5 was endorsed for 3.4% of the boys and for 5.2% of the girls, and item 110 was endorsed for 1.0% of the boys and for 1.7% of the girls (van Beijsterveldt et al. 2006).

With regard to the prevalence of childhood gender dysphoria, information is available from one study. Coolidge et al. (2002) studied the heritability of gtd in a child- and adolescent sample of 96 monozygotic and 61 dizygotic twin pairs (age range 4-17). Based on the information on gtd symptomatology in these twins, assessed through a questionnaire in their parents, a 2.3% prevalence rate was estimated. However, since the study incorporated both children and adolescents, this prevalence rate is likely to be an underestimation of the true prevalence of childhood gender dysphoria. Namely, for most children the gender dysphoria will remit after puberty (see paragraph on development of gender variant and gender dysphoric children) and not all gender dysphoric adolescents have a history of childhood gender dysphoria (e.g. Zucker et al. 2012).

As for the prevalence of gender dysphoria in (later) adolescence and adulthood, studies based on clinical samples indicate that gender dysphoria is relatively rare (approximately 1:7,400 to 1:100,000 in natal males and 1:30,400 to 1:400,000 in natal females; see for overview Zucker and Lawrence 2009). Again, these numbers are likely to be an underestimation of the true prevalence (de Vries and Cohen-Kettenis 2012; Olyslager and Conway 2007; Reed et al. 2009; Wood et al. 2013; Zucker et al. 2008), since most studies based their prevalence estimation on referrals to specialized gender identity services and did not incorporate individuals who experience less extreme forms of gender dysphoria and therefore are not likely to apply for medical interventions (see for an overview Cohen-Kettenis and Pfäfflin 2010). A recent population based study in the Netherlands supports this assumption. It showed that 4.6% of the males and 3.2% of the females in a sample of 8664 participants experienced ambivalence in gender identity and 1.1% of the males and 0.8% of the females reported to have feelings of gender incongruence. Such feelings were not always accompanied with a
desire for (some form of) gender reassignment (hormones and/or surgery), which was reported by 0.6% of the males and 0.2% of the females (Kuyper 2012).

Development of gender variant and gender dysphoric children
At present, knowledge on the psychosexual development of non-normative children consists of the findings from prospective follow-up studies of clinically referred gender dysphoric children and retrospective findings for individuals with a sexual orientation directed towards the same natal sex.

Retrospective studies in homosexual and lesbian individuals show that they recalled more gender variant behaviors in childhood, compared to their heterosexual counterparts (see for an overview, Bailey and Zucker 1995; Zucker et al. 2006). Prospective studies on gender dysphoric children primarily focused on the gender identity / gender dysphoria and sexual orientation outcome in (late) adolescence and (young) adulthood. Collectively, these studies reported on 246 gender dysphoric children, for whom the majority would meet the DSM-IV-TR criteria for a GID diagnosis. In the majority of these children (207, 84.2%) the gender dysphoria remitted after puberty. The findings on sexual orientation outcome in adolescence or adulthood showed an increased prevalence for a sexual orientation directed towards the same natal sex or to both sexes for gender dysphoric children, as compared to gender normative children (Bakwin 1968; Davenport 1986; Drummond et al. 2008; Green 1987; Kosky 1987; Lebovitz 1972; Money and Ruso 1979; Wallien and Cohen-Kettenis 2008; Zucker and Bradley 1995; Zuger 1984).

The understanding of the processes and factors that may be predictive of whether a gender dysphoric child will be a persister (the gender dysphoric feelings persist into adolescence or adulthood), or a desister (the gender dysphoric feelings remit after puberty), or of its future sexual orientation, regardless of being a persister or desister, is limited. One prospective follow-up study by Wallien and Cohen-Kettenis (2008) on 77 clinically referred children (21 persisters, 56 desisters) showed that the percentage of girls with persisting gender dysphoria was higher (50%) than the percentage of boys with persisting gender dysphoria (20%). In addition to this, they found that the cross-gender behaviors, interests and preferences, and the gender dysphoria in childhood were more extreme in the persisters than in the desisters. The results from a retrospective study of 25 gender dysphoric girls, which showed that girls with persisting gender dysphoria recalled significantly more childhood cross-gender behaviors and gender dysphoria than the girls who were classified as desisters, are in line with these findings (Drummond et al. 2008).
Clinical management of gender dysphoric children

At the Center of Expertise on Gender Dysphoria at the VU University Medical Center in Amsterdam, the Netherlands, approximately 50 gender dysphoric children (<12 years of age) are referred each year. Since the start of the clinic in 1987, which was in its early years based at the University Medical Center Utrecht, up to 2012, a total of 468 children, twice as many boys as girls (313 vs. 155) were referred.

In the Dutch clinic the clinical management starts with a diagnostic period, consisting of several sessions with the child, both parents, and the parents individually. The child and the parents are interviewed, and the child is interviewed and observed, and has a psychodiagnostic assessment. Information is also gathered from teachers. The diagnostic procedure aims to determine the intensity of the gender dysphoria. Information on emotional and cognitive, as well as social-, school and family functioning, are assessed in order to establish a potential relationship with the gender dysphoria and seek targets for intervention (de Vries and Cohen-Kettenis 2012).

Vignette: Sue

“Life ain’t easy for a boy named Sue”

(Johnny Cash)

Sue is a 9 year old girl who entered our clinic with her parents on a sunny Wednesday afternoon in June. Dressed in sporty clothing, wearing shiny white sneakers and a short hairstyle, she had the appearance of a boy. According to her parents, Sue always had a greater interest in playing with the cars and action figures of her brother Benny, than playing with the dolls that were given to her for her birthdays. Sue had always disliked wearing feminine clothing. At the first day in kindergarten when Sue was 3 years of age she was in tears and inconsolable, when she had to wear a blue and white dotted dress. This was also the first time that Sue stated that she wanted to be a boy; something she continued to state. Besides these statements, Sue occasionally asked her parents when her penis would start to grow and mentioned that she did not want to become a woman and have breasts in the future. In kindergarten, and later on in primary school, Sue always played with the boys. She had always been an active child; enjoyed climbing trees, playing ball games and had no interests in participating in games with the girls in her class. Sue’s parents did not mind that the behaviors, interests and preferences of their daughter
were different from the other girls. However, since a few months prior, they were worried about Sue’s deteriorating school results, and regular periods of her being sad or even depressed. After a conflict at school about school camp, when Sue said that she would rather die than sleep in the room with the other girls, instead of with the boys, her parents contacted their family doctor, who referred her to our clinic.

Our clinical interventions in gender dysphoric children are not directly aimed to change the child’s gender identity or to suppress or “resolve” their cross-gender behaviors (e.g. de Vries and Cohen-Kettenis 2012), as such treatment attempts in the past have not shown to be effective (see for an overview, Byne et al. 2012). Instead, children often seem to become distressed if their preferences and/or behaviors are blocked rather than channeled (Richardson 1999). Such interventions are now even referred to as unethical (Coleman et al. 2012). Furthermore, clinical associations, such as the American Academy of Child & Adolescent Psychiatry, have formulated their position against interventions that aim to treat gender non-conforming behaviors (Adelson et al. 2012).

In our clinic, interventions focus more on the co-existing problems of the child and the family (if they exist), which may or may not be associated with the child’s gender dysphoria. If counseling is indicated, it is mainly focused on the parents. Topics are addressed, such as: 1.) How to deal with the uncertainty regarding their child’s psychosexual outcome, 2.) How to inform other people about the child’s gender dysphoria and when, 3.) Should the child socially transition to the experienced gender role, and/or 4.) How to handle peer rejection or social ostracism. Medical interventions are not provided in our clinic in childhood and before puberty.

As for the clinical management of gender dysphoric adolescents, defined as youth above the age of 12, medical interventions may be offered in our clinic. However, before any medical interventions are considered, the adolescents go through a diagnostic phase. Similar to the procedure in children, the focus in this first phase is to obtain information on the gender dysphoria, and the psychosocial and family functioning and to assess possible co-existing problems. In addition to this, topics that are relevant in adolescent development are addressed and discussed, such as sexuality and the experience or the anticipation of pubertal body changes. If adolescents fulfill the criteria for a GID diagnosis, they may be eligible for puberty suppression by means of Gonadotropin-Releasing hormone (GnRH) analogues. GnRH analogues suppress the development of secondary sex characteristics. By
putting a halt on this development we “buy time” for the adolescent, so that they are able to further explore their gender identity and their wish for gender reassignment, without the accompanying distress caused by the physical changes of puberty. Puberty suppression is a fully reversible treatment, in that the gonadal function is activated soon after stopping the treatment with GnRH analogues (Mul and Hughes 2008). Beside a GID diagnosis, eligibility criteria for starting with puberty suppression are pubertal (Tanner) stage 2 to 3 and 12 years of age or older, an early onset gender dysphoria that has intensified during the early pubertal phases, an absence of psychosocial or psychiatric problems that may interfere with the diagnostic process or treatment, sufficient support by family or other caregivers, and the ability to understand the impact of a gender reassignment on his or her life. If adolescents experience cross-gender identification and still desire a gender reassignment when they have reached the age of 16, the first step of gender reassignment (cross-sex hormone treatment) may be considered. Eligibility for this treatment phase depends on the same criteria as for puberty suppression (except for the age and Tanner stage criteria). During this phase, where the cross-sex hormones induce the puberty effects of the experienced sex (feminization in natal boys and masculinization in natal girls), adolescents are expected to socially transition. Most of the adolescents, however, will have already made their social gender role change during the period of puberty suppression. When the adolescents reach the age of 18 and have passed through the previous phases successfully and the desire for gender reassignment is still present, they are eligible for surgical interventions. (Cohen-Kettenis et al. 2011; de Vries and Cohen-Kettenis 2012).

**Aims of the thesis**

The aims of this thesis are to enhance our understanding of the development of gender variant and gender dysphoric children and adolescents, and to identify factors associated with the persistence and desistence of gender dysphoria. In order to attain this goal, we studied the psychosexual outcome of gender variant children in the general population and the developmental trajectories of gender dysphoric children. We also investigated possible predictive factors for the persistence and desistence of childhood gender dysphoria and compared children and adolescents referred to two different clinics, one in the Netherlands, and one in Canada.
Outline of the thesis

Gender identity development
Chapter 2 provides a historical overview of the concept of gender identity and describes normative and gender variant gender identity development. Furthermore, possible psychosocial and biological factors contributing to a gender variant identity are described and discussed.

Measuring gender dysphoria in adolescence
Chapter 3 reports on the validity of a 12-item dimensional scale that aims to measure gender dysphoria; the Utrecht Gender Dysphoria Scale (UGDS). The UGDS was administered in a total of 1119 participants; 648 gender dysphoric adolescent and adult participants, 60 participants with a disorder of sex development (DSD), and 219 non-gender dysphoric heterosexual and 192 non-gender dysphoric homosexual/lesbian or bisexual controls.

Psychosexual development and outcome in gender variant and gender dysphoric children
Chapter 4 investigates the association between childhood gender variance and sexual orientation and gender discomfort in adulthood in the general population. In 406 boys and 473 girls the intensity and presence of gender variance was assessed in childhood. Twenty-four years later these children were followed-up in adulthood. Information was gathered on their sexual orientation and current feelings of gender discomfort/gender dysphoria.

Chapter 5 describes a qualitative study which aimed to obtain a better understanding of the persistence and desistence of childhood gender dysphoria and the psychosexual outcome of gender dysphoric children. Twenty-five gender dysphoric children, who met the criteria of a Gender Identity Disorder (DSM-IV or DSM-IV-TR) in childhood, were contacted and interviewed (14 persisters, 11 desisters) when they were on average 16 years of age.

Chapter 6 covers the findings from a quantitative follow-up study on 127 adolescents (47 persisters and 80 desisters), who were referred and diagnosed in childhood (<12 years of age) in the Center of Expertise on Gender Dysphoria, and followed up in adolescence at an average age of 16 years. We examined differences between persisters and desisters in childhood with regard to demographics, developmental background (e.g. birth weight, pregnancy duration), childhood psychological functioning, the quality of peer relations, and childhood gender dysphoria, and adolescent reports of gender dysphoria, body image and sexual orientation.
Chapter 7 represents a short communication where we address the observations in our clinic with regard to the topic of gender transitioning in early childhood and discuss its clinical implications.

Cross-national differences between gender dysphoric children and adolescents

Chapter 8 describes a study that examined the psychological functioning and the quality of peer relations, measured by Teacher report, in 728 clinically-referred gender dysphoric children and adolescents from Toronto, Canada and Amsterdam, The Netherlands. The focus was not only on differences between ages, but also on differences between cultures in order to gain insight in the relationship between gender dysphoria and emotional and behavioral problems.

Discussion and summary

In Chapter 9 the studies are summarized and discussed. Suggestions for future studies and clinical implications are given.