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GENDER IDENTITY DEVELOPMENT IN CHILDREN WITH HEIGHTENED PRENATAL ANDROGEN EXPOSURE

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Abstract

Understanding how gender identity develops has important theoretical implications for typically developing populations and clinical implications for those with gender variability or dysphoria (e.g. low contentment with one's assigned gender) and practical implications for the assignment and management of gender in children born with ambiguous sex/genitalia. While chromosomal females exposed to heightened levels of androgens prenatally consistently express later masculinised/defeminised gendered behaviour, the relationship between prenatal androgen exposure and gender identity development (masculinisation/defeminisation and feelings associated with this) is less consistent. Between-group differences in *gender identity*, *gendered behaviour*, *gender typicality*, *gender contentedness*, and *felt pressure for gender conformity* were examined in a sample of children (age 7 – 11) with *congenital adrenal hyperplasia* (CAH; 23 males, 26 females) together with unaffected siblings (19 males, 29 females). Analyses reveal reduced gender typicality and gender contentedness in girls with CAH together with reduced female gender identity and increased cross-sex gendered behaviour. Bootstrapping mediation analysis examining gender typicality, gender contentedness, and gendered behaviour as mediators between prenatal androgen exposure typicality and gender identity revealed two important relationships: (1) gendered behaviour mediated the relationship between prenatal androgen exposure typicality and gender identity and, (2) this mediation was moderated by gender typicality and gender contentedness. These findings suggest that, for girls with CAH, the increased expression of cross-sex gendered behaviour contributes to the development of desires to be the other sex and that this relationship may depend upon feelings of gender typicality and contentedness.

Keywords: gender identity, gender development, gendered behaviour, congenital adrenal hyperplasia, androgens

Preface

This dissertation is the result of my own work and includes nothing which is the outcome of work done in collaboration except where specifically indicated in the text.

This dissertation does not exceed the word limit for the Department of Psychology.

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INTRODUCTION

Chromosomal females exposed to heightened prenatal androgens consistently display higher rates of masculinised/defeminised gendered behaviour (Hines, 2011a). However, of 14 studies investigating the impact of heightened prenatal androgen exposure in females on gender identity, 9 studies found support for masculinised gender identification (Berenbaum and Bailey, 2003; Gupta et al., 2006; Hines, Brook, and Conway, 2004; Hurtig & Rosenthal, 1987; Meyer-Bahlburg, Dolezal, Baker, Ehrhardt, & New, 2006; Slijper, 1984; Slijper et al., 1998; Woelfle et al., 2002; Zucker et al., 1996) while 5 did not (Dittmann et al., 1990; Ehrhardt and Baker, 1974; McGuire, Ryan, and Gilbert, 1975; Matilla, Fagerholm, Santilla, Miettinen, & Tasinen., 2013; Meyer-Bahlburg et al., 2004). However, the extent to which one can draw conclusions regarding the influence of prenatal androgen exposure on gender identity development based on these previous examinations are limited due to methodological inconsistencies. Case reports and studies investigating the link between prenatal androgen exposure and gender in females suggest that variable or cross-sex gender expression develops separately from (1) incongruent neonatal genital appearance compared to assigned sex (Berenbaum and Bailey, 2003), (2) post-natal socialisation by parents (Pasterski et al., 2005), and (3) relative dose of androgens exposed to prenatally (Speiser, 2001; Wedell, Thilen, Ritzen, Stengler, & Luthman, 1994). This suggests that factors other than those listed above may also be important for the development of typical gender identity. As a result, the development of gender and gender identity is increasingly being understood as a product of the complex interaction between biological, social and cognitive factors.

Prenatal androgen exposure, while responsible for physiological masculinisation, also masculinises and defeminises sexual behaviour in animals (e.g. Beatty, 1992; De Vries & Simerly, 2002; Goy & McEwan, 1980). Additionally, prenatal androgen exposure has been used to manipulate non-sexual but sexually dimorphic animal behaviour. For example, treating female rats and monkeys with testosterone increases subsequent male-typical rough and tumble play (Goy & McEwen, 1980; Meaney & Stewart, 1981). Effects of prenatal androgen exposure on neurobehavioural sexual differentiation has been extensively replicated in many non-human animal species (for a review, see Hines, 2011a). Findings of this nature have driven hypotheses regarding the influence of prenatal androgen exposure on human psychosexual development. However, given ethical concerns regarding hormone manipulation in humans, true experiments examining the influences of androgens on human development are generally not possible. One way to examine these effects then is to study *experiments of nature* - situations in which variant androgen exposure has occurred for other reasons.

There are two disorders of sexual development (DSDs) that occur at a frequency high enough that they act as primary models for understanding the role androgens play on gender development in humans. The first is androgen insensitivity syndrome (AIS). This condition mutates androgen receptors in the brain and body of affected individuals, resulting in subsequent androgen insensitivity. The partial or complete insensitivity to androgens leads to varying degrees of demasculinisation/feminisation in 46,XY individuals (Boehmer et al., 2001). Demasculinisation/feminisation in this population ranges phenotypically from less extreme forms (male infertility or undervirilisation) to more extreme/complete forms characterised by the presence of testes but absence of the Wolffian and Mullerian duct systems and absence of male-typical primary (external genitalia) and secondary sex characteristics. With few exceptions, 46,XY individuals with complete androgen insensitivity are assigned and reared as females and go on to self-identify as female, satisfied with the adoption of the female gender role (Deeb, Mason, Lee, & Hughes, 2005; Hooper et al., 2004), despite infertility. Decisions regarding gender assignment in those with partial androgen insensitivity remain more variable and tend to be based on a number of factors including degree of virilisation, necessity of surgical intervention, possibility of fertility, age of diagnosis and treatment, and parent/patient preference. While gender management remains challenging in this population, studies of AIS provide evidence for the influence of androgens and genes on later gender identity (Wisniewski et al, 2000; Hines, 2004; Hughes, Houk, Ahmed, & Lee, 2006).

The second condition which acts as a primary model for understanding the role of prenatal androgens on gender development, and the focus of this analysis, is classical congenital adrenal hyperplasia.

Classical Congenital Adrenal Hyperplasia

Classical congenital adrenal hyperplasia (CAH) is an autosomal recessive condition that causes overproduction of adrenal androgens due to impaired cortisol synthesis (Speiser et al., 2010). Presenting in early gestation, CAH occurs in 1:10,000-20,000 births (Merke & Bornstein, 2005). 95% of cases are caused by mutations in the CYP21A2 gene leading to deficiency of 21-hydroxylase (21-OH) the enzyme responsible for converting 17-hydroxyprogesterone (17-OHP) to 11-deoxycortisol and progesterone to deoxycorticosterone, respective precursors for cortisol and aldosterone synthesis (Krone, Dhir, Ivison, & Arlt, 2007; White & Speiser, 2000). This deficiency results in one of two classical forms of the condition: (1) the salt-wasting (SW), or severe form of CAH whereby the synthesis of cortisol (later converted to androgens) and aldosterone are markedly to fully inhibited, or (2) the simple-virilising (SV), or mild form, presenting less cortisol and reciprocal androgen inhibition. There is an additional non-classical form of the condition which results in the onset of impaired cortisol synthesis later in life and is typically less severe than the classical forms. In the classical forms of CAH, the focus of this paper, diagnosis and treatment usually occurs in 46,XY infants following life-threatening salt and water loss and high serum potassium levels, and in 46,XX infants following the presentation of genital virilisation or ambiguity. For these reasons, diagnosis usually occurs earlier in 46,XX infants than in 46,XY infants (Speiser et al., 2010). Immediate treatment with glucocorticoids and mineralocorticiods attempts to restore balance to the endocrine environment and is generally effective.

CAH and Psychosexual Development

Research examining human psychosexuality (gendered behaviour, gender identity, and sexual orientation) in individuals with CAH reveals that females display a marked shift toward the male-typical compared to female controls in some important ways. The gendered behaviour of girls with CAH shows a well-established shift toward the male-typical in toy, playmate, and activity preferences compared to unaffected female relatives or matched controls (Berenbaum & Hines, 1992; Dittmann et al., 1990, Ehrhardt & Baker, 1974; Ehrhardt, Epstein, & Money, 1968; Hall et al., 2004; Hines, Brook, & Conway, 2004; Meyer-Bahlburg et al., 2004; Nordenstrom, Servin, Bohlin, Larsson, & Wedell, 2002; Pasterski et al., 2005, 2007, 2011). In adulthood, females with CAH show a shift toward male-typical erotic target preference with increased levels of non-heterosexuality compared to unaffected female relatives or matched controls (Frisen et al., 2009; Hines, 2011b; Meyer-Bahlburg et al., 2008; Zucker et al., 1996).

As mentioned previously, the same consistency observed regarding the influence of prenatal androgen exposure on gendered behaviour and sexual orientation in females is not observed with regard to gender identity outcomes. While findings appear more mixed, a review of previous studies examining gender identity in female CAH populations (Berenbaum and Bailey, 2003; Dittmann, et al., 1990; Ehrhardt and Baker, 1974; Gupta, et al., 2006; Hines, Brook, and Conway, 2004; McGuire, Ryan, and Gilbert, 1975; Meyer-Bahlburg, et al., 2004; Meyer-

Bahlburg, et al., 2006; Slijper, 1984; Slijper, et al., 1998; Woelfle, et al., 2002; Zucker, et al., 1996) reveal that most (9 of 14) observe a reduction in female gender identity in girls and women with CAH compared to female controls. Further, more women with CAH who are reared as girls, compared to women with normative levels of prenatal androgen exposure, later change to live as men (Dessens, Slijper, & Drop, 2005). Increased variability in gender identity in females with CAH suggests that the factors influencing the development of gender identity may be more varied or different than factors influencing the development of gendered behaviour. While prenatal androgen exposure is consistently linked to the masculinisation of gendered behaviour in females with CAH, the role other factors may play in development of gender identity may account for inconsistencies in the link between prenatal androgen exposure and gender identity. For example, biological, social, cognitive, and affective factors suspected and demonstrated as important to the development of gender could influence gender identity directly (like in the case of the direct link between prenatal androgen exposure and gendered behaviour) or indirectly, via other mechanisms or involving an interaction of multiple factors which then result in the development and expression of typical or variable gender identity (Maccoby, 2000). Understanding these factors and the role they play in the development of gender identity stands to aid the clinical care of female patients with CAH as well as elucidate processes involved in gender identity development more generally.

CAH and Gender Identity

Gender identity typically refers to the sense of self an individual has as being male or female (Hines, 2004). For most, one's gender identity coincides with one's biological sex. However, for some, an incongruence may be experienced which, in extreme cases, can be associated with or result in an intense and persistent desire to be the other sex (Zucker & Lawrence, 2009). These individuals are thought to be gender incongruent or gender variant. Gender variance is typically conceptualized as residing along a continuum with normative gender expression on one end and more severe gender variance and possible accompanying gender dysphoria (cross-sex ideation and the distress associated with these ideations) on the other. The conceptualisation of this experience by the American Psychological Association (APA) was previously Gender Identity Disorder (GID) in the DSM-IV-TR (2000) for those without a DSD, now Gender Dysphoria (GD) in the DSM-5 (2013) for those with and without a DSD. Generally, GID/GD are characterised and assessed by the presentation of cross-sex ideation (e.g. desire to be the other sex), cross-sex behaviour, the dislike for the attributes of one's own sex, and the preference for attributes of the other sex. In a sample of 250 girls with CAH, 5.2% experienced gender problems, 2.8% experienced gender dysphoria (discontent with assigned gender), and 1.6% changed to live as men in adulthood (Dessens, Slijper, & Drop 2005). While the rate of gender change in females with CAH is higher than expected in the general population (Meyer-Bahlburg et al., 1996; Sripathy, Ahmed, Sakati, & al-Ashwal, 1997; Zucker et al., 1996), the masculinisation/defeminisation of gender identity appears to be less than the observed masculinisation/defeminisation of gendered behaviour and most (94.7%) 46,XX females reared as girls grow up satisfied with their assigned gender (Dessens, Slijper, & Drop, 2005).

Despite these conceptualisations, gender identity is increasingly being viewed and measured as separate from gendered behaviour (Zucker, 2010). Pasterski et al. (2014) present a detailed examination of gender identity and gendered behaviour in children with CAH and find support for the distinctness of the two constructs. Specifically, while the gender identity and gendered behaviour correlated overall, they observed that this correlation was driven by scores of the CAH girls and was not significant in the other groups of children due to a lack of variance in scores (e.g. SD = 0.00 for the mean gender identity score for boys with CAH). Further, age negatively correlated with gender identity but positively correlated with gendered behaviour. Previous quantifications of gender identity and, specifically, the inclusion of gendered behaviour in its assessment, may therefore be one potential source of the inconsistency in findings examining the link between prenatal androgen exposure and later gender identity. While one's gender identity and gendered behaviour are typically examined simultaneously for GID/GD diagnoses and while these two facets typically correlate, it is clear that this may not always be the case (Dessens, Slijper, & Drop, 2005). As such, previous examinations of gender identity shifts in females with CAH may have confounded, at least to some degree, gender identity and gendered behaviour. Our increasing understanding of the various factors associated with gender and gender identity, how these factors relate to one another, and how gender identity should be quantified is ever progressing, a progression signified by changes in the conceptualisation of GD in the DSM-5. With these changes in mind together with findings presented by Pasterski et al. (2014), it is possible that a number of the studies listed above have used measures (or, in some cases, single items) that may have had weak construct validity, resulting in a blurring of the ability to observe the direct or indirect roles prenatal androgens might play in gender identity development. In addition, this may have limited the ability to compare findings between studies.

The practical difficulties of working with a rare condition like CAH further weaken construct validity. Specifically, acquiring a sample of sufficient size to ensure strong statistical power for testing these hypotheses can be particularly challenging. This is the case for a number of the reviewed studies examining prenatal androgen exposure and gender identity. Sample sizes tend to be small or lacking appropriate control groups, methodological concerns which increase the likelihood of Type 2 error. In addition, age is an important factor when examining CAH and gender identity due to the relatively high prevalence of desistance of GID from childhood to adulthood (Green, 1987; Singh, 2012; Wallien & Cotton-Kettenis, 2008). Yet, the scores of both children and adults were combined for a number of the reviewed studies. Finally, both self- and parent-reported information is often used when working with children despite evidence suggesting that data can differ in important ways depending on the sources of the information (De Los Reyes & Kazdin, 2004). In sum, small sample sizes, age variability, and different sources of information all present limitations of previous studies examining prenatal androgen exposure and gender identity. Subsequent examinations should make efforts to minimise the impact of these limitations on the conclusions being drawn.

Gender Identity Development

Gender is an important component of human identity and, as such, has been the focus of much research. In general, while the biology, environment, and cognitions of males and females are more similar than different, they also differ in important ways. Contemporary research has therefore spent considerable energy trying to understand how each of these components relates to one of the strongest sex differences observed in humans, gender identity (Hines, 2004). Early explanations for gender differences focused on the influence of direct socialisation (see Maccoby 2000 for a review of perspectives of gender development). Here it was thought that children take on the attributes appropriate for their own sex as a function of positive and negative reinforcement exerted by socialisation agents (parents, teachers, peers, etc.). The role of the child in the process was viewed largely as passive and both within- and between-sex differences were thought to reflect the same process of reinforcement. This perspective was later elaborated upon to include the influence of indirect socialisation via modelling and imitation. Here, it was thought that in addition to the direct impact of reinforcement, children take more active roles in their own gender development by observing and imitating same-sex others. It became clear, however, that these processes necessitated personal and external gender membership knowledge together with the ability to summarise the same- and other-sex patterns of others and the motivation to adopt same-sex patterns while rejecting other-sex patterns. As such, the role of cognition on gender development became apparent. Here, the child was seen as active selectors and users of pertinent and developmentally appropriate gender information and formed gender schemas, cognitive structures that organise knowledge into sets of expectations that guide and organise social perceptions (Bem, 1981).

Support for the influence of social and cognitive processes on gender identity development comes from a wide range of research. Gender is one of the first parts of identity that is developed and expressed, it's formation usually taking place in steps (Slaby & Frey, 1975), characterised by simultaneous processes of individual identification, categorisation, and comparison. For example, by the age of 2.5 – 3 years, knowledge of one's gender membership solidifies (Ruble et al., 2007). From the age of 3 years, sex segregation begins whereby same-sex playmates are preferred (Maccoby & Jacklin, 1987), a process observed in a variety of situations and across a diverse array of cultural and social environments. Between the ages of 4 and 6 years, gender role establishment occurs whereby a "trying on" of roles is undergone through natural, gendered and cross-gendered fantasy and play. By the age of 6 – 7 years, it is usually understood that one's gender will remain constant across time despite variable appearance (Martin & Ruble, 2004). Finally, around the age of 6 – 8 years, gender-stereotyping begins to grow more flexible (Ruble et al., 2007). As such, children begin to understand that violations to the typical gender role for their specific society and zeitgeist (time in history) do not necessarily represent different gender identifications.

While it is clear that socialisation and cognitions play a role in the development of gender, more recent evidence regarding the influence of biological factors has further enhanced our understanding of the development of gender in humans. For example, research has demonstrated that before a child outwardly expresses or identifies with one particular gender over another, sex-differentiated behaviour is expressed. Connellan, Baron-Cohen, Wheelwright, Batki, and Ahluwalia (2000) found that infant females of 12 months of age or younger, preferentially directed eye-gaze toward faces while infant males directed their gaze toward the physical-mechanical. Differences like these are hypothesised to represent early expressions of gender and appear to occur too early to be best explained by socialisation or cognitive processes. As stated earlier, studies examining sex-differentiated behaviour in non-human animals further support the role biology plays in the development of sex-differentiated behaviour. As such, it is clear that there are social, cognitive, and biological features to the development of gender. Further, it is becoming equally clear that each of these factors likely influences gender at varying times throughout development and to varying degrees. As a result, the thinking regarding gender development has shifted, emphasising the importance of both critical periods for gender development (Hines, 2004) as well as the role the *interplay* of these processes have on development (Maccoby, 2000).

Before engaging in a discussion of what gender identity-related factors might influence one's gender identity and how, there are a few points warranting distinction. First, gender differentiation shows demonstrable within-sex variability. When wide, overlapping per sex distributions exist for a particular characteristic, it is still possible, and even probable, that some females demonstrate higher levels of a male-typical characteristic than some males and vice versa. Second, gender differentiation shows demonstrable cultural and temporal variability. In other words, the male- or female-typicality of a particular characteristic can range from culture to culture and can change over time. Finally, gender differentiation, which is increasingly viewed as consisting of various domains (e.g. activities, interests, traits), can show within-individual variability (Egan & Perry, 2001). An individual's gender typicality on one domain does not necessarily reflect their typicality on other domains. Further, this within-individual variability can be observed in the sex-typing of the self (how they perceive themselves as congruent with their gender) compared to the sex-typing of the other (how they perceive others as congruent with their gender). Each of these points must be taken into consideration when interpreting the results of all examinations of gender in humans.

With these distinctions in mind, we can begin to examine how a child's sense of gender congruency (or incongruency) might influence their gender identification and their satisfaction with this identification. With gender membership and constancy knowledge in tact together with increased flexibility in gender-stereotyping, children begin to perceive and identify elements of their gender on a number of domains as consistent with prescribed gender roles or not, eventually working towards an overall appraisal of their own gender. In other words, their overall gender identity. However, unifactorial conceptualisations of gender identity are increasingly being viewed as inadequate or misleading (Yunger, Carver, & Perry, 2004). This is because such conceptualizations may ignore the interactive influence of many gender-related factors on gender identity outcomes. As a result, gender identity and its formation has been increasingly viewed as multifactorial (Egan & Perry, 2001; Spence, 1993). In other words, gender identity can be thought of as the composite of or one's feelings of compatibility with, motivation to fit in with, and evaluation of their gender. Egan and Perry (2001) therefore identified five factors important in understanding how gender identity develops (and how it relates to mental health). These factors include (a) *membership knowledge* (knowledge of one's membership in a gender category); (b) *gender typicality* (perceived sense of similarity to others of one's own gender together with a perceived dissimilarity to others of the opposite gender); (c) *gender contentedness* (satisfaction with one's gender assignment); (d) *felt pressure for gender conformity* (pressure felt from parents, peers, and the self to conform to be similar to those of one's gender and dissimilar to those of the opposite gender); and (e) *intergroup bias* (the belief that one's own sex is superior to the other).

Egan and Perry (2001) examined the relationship between these gender identity-related factors and adjustment (*global self-worth, social competence,* and *peer acceptance*) in a large sample of US children (age 8 - 12) and their findings supported the conceptualisation of gender identity as multifactorial. Further, they found that gender compatibility (quantified as self-perceived *gender typicality* and the contentment derived from this typicality, *gender contentment*) was positively associated with adjustment, whereas felt pressure and intergroup bias were negatively associated with adjustment. Specifically, gender typicality was positively related to all measures of adjustment and neither gender contentedness nor felt pressure for gender conformity better explained this relationship. An *a priori* prediction also led researchers to find that the degree to which gender contentedness related to self-worth was a direct function of the degree to which children felt pressure to conform to gender stereotypes – in other words, low gender contentedness was harmful to self-worth only in the presence of high felt pressure for gender conformity. Egan and Perry (2001) concluded that it was felt pressure for gender conformity that was maladaptive to adjustment and that a child's gender identity benefits from a secure conception of themselves as a typical member of their own sex together with the freedom to explore cross-sex options when they desire to do so.

Hypotheses

Given the results presented by Egan and Perry (2001), it was hypothesised that the presented gender identity-related factors might also play a role in the development of gender-related adjustment or, more specifically, to gender identity. How girls with CAH self-assess and perceive their own gender as typical or not and the degree of satisfaction or contentment derived from this appraisal has not been previously examined. Further, if these factors contribute to overall adjustment, it is reasonable to suspect that they they might also contribute to gender-specific adjustment, or, more specifically, gender ideations (ranging from same- to cross-sex ideations). As such, the gender identity-related factors proposed by Egan and Perry (2001; *gender typicality, gender contentedness,* and *felt pressure for gender conformity*) were measured in a population of children with CAH and their unaffected siblings with the goal of ascertaining the role these factors might play in the development of gender identity. Given the ability to measure intergroup bias was greatly diminished by the rarity of CAH, this measure was not included. Further, gender identity was examined separately from gendered behaviour given evidence suggestion the distinctness of these constructs and in order to determine the role gendered behaviour might play specifically in the development of gender identification and ideation (Pasterski et al. 2014, Zucker, 2010).

As previously reviewed, it is hypothesised that girls with CAH will demonstrate less sex-typed gendered behaviour and more cross-sex gender ideation compared to boys and unaffected girls. Based on the results of Egan & Perry's (2001) analysis, it was hypothesized that girls will feel less gender typical and less gender content than males while males will feel more pressure for gender conformity. As such, I predict that boys with CAH and their unaffected male counterparts would feel more gender typical than girls with CAH and their unaffected female counterparts with girls with CAH feeling the least gender typical. Additionally, I predict that both boys with CAH and their unaffected male counterparts will feel more gender content than girls with CAH and their unaffected female counterparts with girls with CAH feeling the least gender content. I also predict that boys with CAH and their unaffected male counterparts will feel more pressure for gender conformity than girls with CAH and their unaffected female counterparts with girls with CAH presenting levels of felt pressure for gender conformity that is shifted in a male-typical direction so that it lies intermediate between the levels felt by boys and unaffected girls. Finally, I predict that gendered behaviour, gender typicality, gender contentedness, and felt pressure for gender conformity will all mediate the relationship between prenatal androgen exposure typicality and gender ideations.

METHOD AND MATERIALS

Participants

Participants included 49 children with classical CAH due to 21-OH deficiency: 26 girls (23 salt-wasting [SW], 3 simple-virilising [SV]) and 23 boys (21 SW, 2 SV). There were 48 controls: 29 females (20 sisters, 9 first cousins); and 19 males (14 brothers, 5 first cousins; see Table 1 for participant age and parent information), all between the age of 7 and 11. 91.7% of participants were Caucasian and recruitment included invitations sent through a national CAH support group and endocrinologists at 11 clinics throughout the UK. All parent participants were fully informed with respect to procedures and consented to their own and their child's inclusion in the study. Assent was provided by each child participant. The recruitment and research protocol was approved by national and institutional research ethics committees and data were stored separately from identifying information. Data stored electronically were password protected and encrypted.

Table 1: Participant Age and Parent-report Information.

		Males		**Females**	
		Unaffected	**CAH**	**Unaffected**	**CAH**
		n = 19	n = 23	n = 29	n = 26
Participant age	**Mean**	9.40	8.71	8.98	8.82
	SD	1.52	1.25	1.68	1.44
	Range	6.99-11.93	7.10-11.34	7.01-11.86	7.02-11.90
Parent reporting	**Mothers**	17	23	28	25
	Fathers	2	0	1	1

Measures

The gender identity-related measures were adapted from the measures used by Egan and Perry (2001). These measures were originally designed to ascertain unique cognitive and affective factors related to childhood gender identity. The items used were variations of items published by Egan and Perry (2001). Therefore, principal components analyses and factor analyses were conducted on each of these measures in order to ensure and increase construct validity. When possible, these analyses also revealed which items best assessed the respective constructs. Those items that best measured the intended constructs were maintained in subsequent analyses. For analyses on these measures, additional principal components analyses and factor analyses (FA) were also conducted separately per sex. These analyses were conducted in order to ensure results from the overall analyses were representative of values depicted per sex. The results of these analyses informed, at least in part, the decision on how to conduct per measure factor analyses – in other words, the results aided in choosing the most appropriate types of extractions and rotation methods used. Where there were indication of sex differences in these preliminary examinations, they appeared to be the result of group differences in condition (unaffected vs. CAH) as opposed to broader sex differences in responding to these measures. As such, overall factor analyses conducted on these measures appear reliable.

For an extensive assessment of childhood gender identity, two parent-report measures (one interview and one questionnaire) and a child self-report interview were administered. These measures were chosen, in part, because they were previously validated or because they reflected widely used methods for the general and clinical examination or assessment of gender identity. Factor analysis was conducted on these measures in order to ensure gender identity was being measured separate from gendered behaviour (see Appendix 1 for items; description of this process is described later).

When possible, the format of each of the measures assumed that which was suggested by Harter (1985), a format which was designed to decrease the effects of socially desirable responding. This format was used for the *gender typicality, gender contentedness,* and *gendered behaviour scales* and details of this method are outlined in the description of these measures (see below). Items of these measures were presented in a mixed order. Each gender identity-related measure (*gender typicality, gender contentedness, felt pressure for gender conformity, gendered behaviour)* was chosen because it, in general, measured a range of gender domains (e.g. interests, feelings, personality). When possible, self-report (as opposed to parent-report) measures were used in an attempt to focus on individual experiences and appraisals and in order to maintain accuracy. Each of these particular measure features was chosen in response to weaknesses of previous examinations on gender identity development.

Gender typicality scale (GT). This 22 item scale was developed to measure the degree to which a child feels that he or she is a typical member of his or her gender. Individual items were worded, for example, as follows: "Some girls have the same interests that other girls have, BUT, Other girls don't have the same interests that other girls have." Instructions asked children (girls in this case) to first indicate which kids they were most like (for example, if they have or don't have the same interests that other girls have). They were then asked to tell if this was "very true" or "sort of" true for them. Initial factor extraction estimation using principal components analysis revealed a two factor solution when examining the Eigen values and Scree plot for these items. Factor analysis using Generalized Least Squares extraction and Varimax rotation methods revealed that these factors accounted for 29.8% and 20.4% of the variance, respectively. Items loading higher than .6 were maintained: Factor 1 labelled "other gender" (5 items) and Factor 2 labelled "same gender" (2 items). Each factor contained items regarding feelings, personality, and interests or activities. Factors loaded dichotomously on the unrotated factor matrix suggesting participants treated these items as unidimensional with masculinity on one end of the scale and femininity on the other (see Appendix 2 for a list of item wording and loadings). As such, the 7 remaining items were treated as one measure. Internal consistency reliability of the 7 items was high (Cronbach's $\alpha = .85$). A unidimensional mean score was computed for each participant with higher scores indicating higher gender typicality.

Gender contentedness scale (GC). This 12 item scale was developed to measure the degree to which a child is happy with his or her assigned gender. Individual items were worded, for example, as follows: "Some girls are happy that they are a girl, BUT, Other girls are not happy that they are a girl." Instructions asked children (girls in this example) to first indicate which kids they were most like (for example, the happy one or the not happy one). They were then asked to tell if this was "very true" or "sort of" true for them. Initial factor extraction estimation using principal components analysis revealed a two factor solution when examining the Eigen values and Scree plot for these items. Factor analysis using Generalized Least Squares extraction and Varimax rotation methods revealed that these factors accounted for 32.8% and 24.3% of the variance, respectively. Items loading higher than .6 were maintained. When examining high loading items, it was discovered that items loading on Factor 2 related much less specifically to gender contentedness (see Appendix 3 for a list of item wording and loadings). These items were therefore discarded for remaining analysis. The remaining 4 items were treated as one measure. Internal consistency reliability of the 4 items was high (Cronbach's $\alpha = .85$). A unidimensional mean score was computed for each participant with higher scores indicating higher gender contentedness.

In order to ensure that the *gender contentedness* and *gender typicality* scales were indeed measuring different constructs, factor analysis was conducted on all remaining, post-factor analysis gender typicality and gender contentedness items (11 in total). Principal components and factor analysis revealed that the remaining gender typicality and gender contentedness items loaded on the 3 factors which would be predicted based upon the previous factor analyses: gender typicality (same gender), gender typicality (other gender), and gender contentedness.

Felt pressure for gender conformity scale (FP). This scale consisted of 24 items and was developed to measure the extent to which a child feels pressure from parents, peers and self, for conformity to gender stereotypes. Individual items were worded, for example, as follows: "My parents want me to be similar to other girls". Instructions asked children to indicate how true they felt the statement was for them using a 4-point Likert scale (1 = not at all, 2 = a little, 3 = pretty true, and 4 = very true). Due to the number of *a priori* factors included in this scale (felt pressure for gender conformity, other gender- vs. same gender-oriented items, and source of pressure [parent, peer, self]), factor analysis of the 24 items remained unhelpful in delineating the most useful items. For example, when one factor was extracted using Generalized Least Squares extraction and Varimax rotation method, only two items loaded with values above .6 and, when two factors were extracted, only 5 items representing random domains loaded at values above .6. Given the impact of the multiple *a priori* factors influencing the results of the factor analyses and given that the internal consistency reliability of the 24 items was high (Cronbach's α = .86), all items were maintained for subsequent analyses. A mean score was computed for each participant with higher scores indicating more felt pressure for gender conformity.

Gendered behaviour using the Children's Activities Inventory (CAI; Golombok & Rust, 1993a; Golombok & Rust, 1993b; Golombok et al., 2008). The CAI is an age-appropriate, modified version of the Pre-School Children's Activities Inventory, a previously validated, self-reported measure of involvement in a variety of sex-typed behaviours for children. The CAI comprises 24 items total, 12 of which are masculine, 12 of which are feminine. Of these items, 7 refer to toy preferences, 11 refer to activity preferences, and 6 refer to characteristics of the child Individual items were worded, for example, as follows: "Some kids play with dolls, BUT, Other kids don't play with dolls." Instructions asked children to first indicate which kids they were most like (for example, the kids who play with dolls or the kids who don't play with dolls). They were then asked to tell if this was "really true" or "sort of" true for them. Mean scores were then standardised with higher scores indicating more masculine behaviour and lower scores indicating more feminine behaviour. Mean standardised scores of the unaffected participants (unaffected girls, M = 39.41, SD = 14.69, unaffected boys, M = 62.55, SD = 6.76) and of the boys with CAH (M = 59.95, SD = 7.11) did not differ significantly from standardised scores obtained from more than 2,000 children in the UK, Netherlands, and US (Golombok & Rust, 1993a; boys, M = 60, SD = 10; girls, M = 40, SD = 10). As such, we can be reasonably assured that these control groups are representative of the general population. For the purposes of subsequent analyses, girls scores were then subtracted from 100 (unaffected girls, M = 60.59, SD = 14.68; girls with CAH, M = 45.98, *SD* = 12.37). By doing this, the measure was converted to a range spanning from cross-sex (lower scores) to sex-congruent gendered behaviour expression with higher scores indicating greater involvement in sex-typed behaviours. This scale will be subsequently referred to as *gendered behaviour* (GB).

Structured diagnostic interview for gender identity disorder (SDIGID). At the time of data collection, the DSM-5 (APA, 2013) was not yet published. As such, the SDIGID was designed to measure DSM-IV-TR diagnostic criteria for GID (APA, 2000). The SDIGID is a parent-report interview assessing a child's cross-sex ideation, cross-sex behaviour, and dislike of one's own, and preference for attributes of the other, sex. Parents were asked to indicate frequency with which the child exhibited each criterion using a 4-point Likert scale (ranging from 1 = frequently to 4 = never). The criteria which requires a stated dislike of one's genitalia were excluded given the sex difference in genital incongruence in this population and the within female sex variability of genital virilisation/surgical manipulation and appearance (Speiser, et al., 2010). Additionally, given the population, the stipulation that each child not have a diagnosis of DSD was disregarded. As per the DSM-5 reconceptualisation (APA, 2013), this is also the current practice for all individuals being considered for a GD diagnosis.

Parent-report Gender Identity Questionnaire for Children (GIQC; Johnson et al., 2004). The GIQC was developed to correspond to the core phenomenology of the cross-sex ideation and behavioural characteristics of GID. This 16-item, previously validated measure uses 12 items on a 5-point Likert response scale (ranging from 1 = every day to 5 = never). An additional 4 items assess the desire to be the other sex and anatomic dysphoria. The sensitivity rate of this measure is 86.8% for a specificity of 95% (Johnson et al, 2004). For the current study, the items pertaining to anatomical dysphoria were disregarded given the sex difference in genital incongruence in this population and the within-female sex variability of genital virilisation/surgical manipulation and appearance (Speiser, et al., 2010).

Gender Identity Interview for Children (GIIC; Zucker, et al., 1993; Wallien, et al., 2009). This measure contains 12 items assessing the child's cognitive and affective understanding of his/her gender including degree of cross-sex ideation. Individual items were worded, for example, as follows: "Are you a boy or a girl?" and "Do you ever feel more like a boy than a girl?" Each item was scored by the interviewer as "2" for a sex-appropriate response, as "1" for an ambiguous response, or as “0” for a cross-sex response. A sum of item scores, ranging from 0 to 24, was then computed.

Analyses of the Gender Identity Questionnaires. Because gender identity and gendered behaviour are increasingly being viewed as independent constructs (Pasterski et al. 2014, Zucker, 2010), analyses were conducted to create composites of the gender identity and gendered behaviour items independently and are described in more detail in Pasterski et al. (2014). Principal components and exploratory factor analysis were performed on the two measures which included both gender identity and gendered behaviour items (i.e., the GIQC and the SDIGID) using Maximum Likelihood extraction and Varimax rotation methods (see Appendix 1 for factor loadings). Items from the GIIC were not included in the factor analysis because all items appeared to pertain directly to identity and not to gendered behaviour and because variance on many of the individual items on the GIIC was low.

As expected, the two identity items from the GIQC and the single identity item from the SDIGID loaded on a single factor. The majority of the remaining items loaded on two other factors. Factor 1 consisted of gender role or gendered behaviour items. Factor 3 included three items which appear to have confounded masculine and feminine interests (i.e., playing sports with girls, playing with a boy-type doll, and imitating male characters). Despite the low variance in the GIIC scores, ANCOVA revealed the expected pattern of results (e.g. significant main effects of sex and condition) with the scores of girls with CAH significantly lower than the scores of the other groups). Therefore, scores for the identity items on each of the two measure were standardised and averaged together with the sum scores from the GIIC to create a *gender identity composite* (GI) with lower scores indicating decreased sex-typicality or increased cross-sex ideations (e.g. increased desire to be the other sex). In response to a weakness of previous examinations of gender identity development in individuals with CAH (e.g. source of information), only 2 non-self-report items were used in these analyses and thus make up only a small part of the gender identity composite.

All analyses were conducted using SPSS Version 21.0. Per group means and standard deviations of each of the measures used in subsequent analyses (GT, GC, FP, GB and GI Composite) are presented in Table 2. Given the previously outlined role of age in gender development, participant age was assessed as a possible covariate. A two-way ANOVA was conducted with participant age entered as the dependent variable and with sex and condition entered as the fixed factors. No significant main effects were observed (sex: $F[1, 96] = 0.272$, $p = .603$, $\eta_p^2 = .003$; condition: $F[1, 96] = 1.893$, $p = .172$, $\eta_p^2 = .02$) nor was the interaction effect significant ($F[1, 93) = .752$, $p = .388$, $\eta_p^2 = .01$). The correlations between age and the measures were also examined per group and are presented in Table 3. Only the age of unaffected girls correlated with gendered behaviour. As such, age was monitored as a significant covariate for all general linear models. In these analyses, age was not found to be significant. Therefore, age was not controlled for in the analyses displayed below.

Table 2: Means and Standard Deviations for Gender Measures.

Measure	Sex	Unaffected			CAH		
		N	Means	SD	N	Means	SD
GT	Boys	19	3.35	.56	23	3.47	.41
	Girls	29	2.85	.55	26	2.26	.84
GC	Boys	19	3.87	.28	23	3.95	.21
	Girls	29	3.75	.37	26	3.32	.86
FP	Boys	19	2.38	.79	23	2.59	.45
	Girls	29	2.09	.55	26	2.10	.41
GB	Boys	19	62.55	6.76	23	59.95	7.11
	Girls	27	60.59	14.69	26	45.98	12.37
GI	Boys	19	.31	.14	23	.33	.10
	Girls	29	.31	.10	26	-.60	1.38

Note: GT – Gender typicality; GC – Gender contentedness; FP – Felt pressure for gender conformity; GI - Gender identity composite – z-scores; GB – Gendered behaviour as per sex-typed CAI scores.

Table 3: Overall and Per Group Correlations between Age and Gender Measures.

Participant age	Overall			
	n	*Pearson's r*		
GT	97	.092		
GC	97	.058		
FP	97	-.103		
GB	95	-.085		
GI	97	.150		
	Unaffected girls		Unaffected boys	
GT	29	-.061	19	-.154
GC	29	.093	19	-.339
FP	29	-.285	19	-.060
GB	27	-.509**	19	-.045
GI	29	-.018	19	-.383
	Girls with CAH		Boys with CAH	

GT	26	.334	23	.101
GC	26	.162	23	-.140
FP	26	.048	23	-.024
GB	26	.271	23	-.031
GI	26	.322	23	.031

* $p < .05$; ** $p < .01$

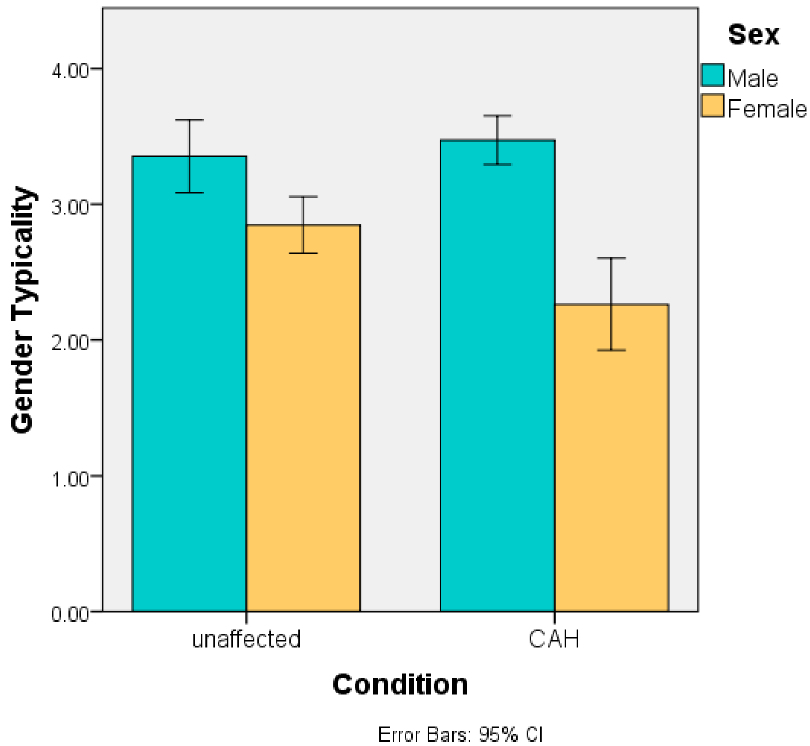
Note: GT – Gender typicality; GC – Gender contentedness; FP – Felt pressure for gender conformity; †GI - Gender identity composite – z-scores assessed; GB – gendered behaviour as per sex-typed CAI scores

RESULTS

Results are presented in three parts. The first includes a series of general linear models (ANOVA's) examining between group differences for each measure including an assessment of main effects, interaction effects, and, when appropriate, post-hoc comparisons. The second includes a brief examination of the correlations among measures. Finally, the relevant gender identity-related factors are assessed as mediators between prenatal androgen exposure typicality (in other words, girls with CAH vs. all others) and gender ideations using the gender identity composite. Results shown are two-tailed.

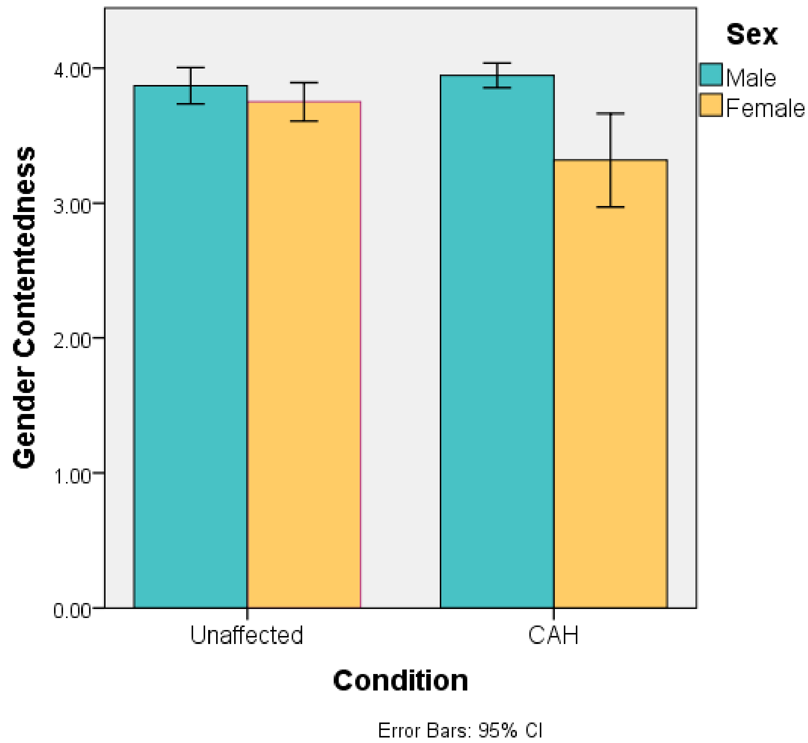
A two-way ANOVA was conducted with GT acting as the dependent variable and with sex and condition acting as the fixed factors. While the main effect of condition trended toward significance (F[1, 96] = 3.37, $p = .069$, $\eta_p^2 = .035$), a significant main effect of sex (F[1, 96] = 45.88, $p < .001$, $\eta_p^2 = .33$) and a significant interaction effect (F[2, 95] = 7.70, $p = .007$, $\eta_p^2 = .076$; see Figure 1) were observed. Subsequent post-hoc pairwise t-tests found that the scores of all groups were significantly different from one another (girls with CAH girls v. unaffected girls: *t*[42.43] = -3.02, $p = .004$; girls with CAH v. boys with CAH: *t*[37.52] = -6.52, $p < .001$; girls with CAH v. unaffected boys: *t*[43] = -4.93, $p < .001$; unaffected girls v. boys with CAH: *t*[50] = -4.53, $p < .000$; unaffected girls v. unaffected boys: *t*[46] = -3.11, $p = .003$) except for the scores of boys with CAH and their unaffected male (*t*[40] = .792, $p = .433$). As such, the scores of the girls with CAH were the lowest, with the scores of the unaffected girls intermediate between the scores of the boys and of the girls with CAH.

Figure 1: Interaction effect for GT.



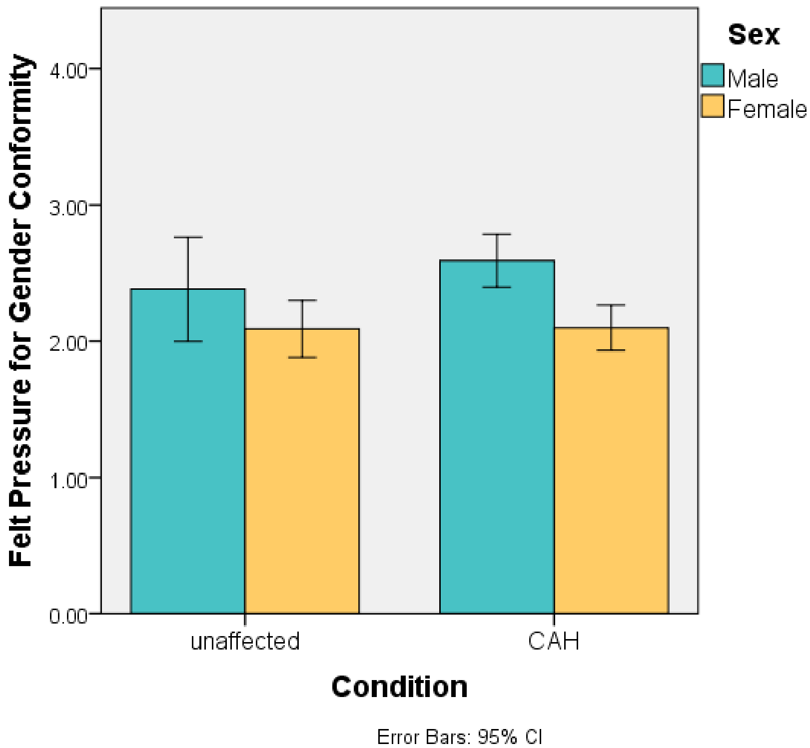
A two-way ANOVA was conducted with GC acting as the dependent variable and with sex and condition acting as the fixed factors. While there was no significant main effect of condition (F[1, 96] = 2.82, p = .096, η_p^2 = .029), a significant main effect of sex (F[1, 96] = 12.46, p = .001, η_p^2 = .118) and a significant interaction effect (F[2, 95] = 5.81, p = .018, η_p^2 = .059; see Figure 2) were observed. Subsequent post-hoc pairwise t-tests found that the scores of girls with CAH were significantly lower than scores of all other groups (unaffected girls: t[33.33] = -2.38, p = .023; unaffected boys: t[31.95] = -3.07, p = .004; boys with CAH: t[28.45] = -3.62, p = .001) while only the scores of unaffected girls were lower than the scores of boys with CAH (t[45.90] = -2.38, p = .021; unaffected girls v. unaffected boys: t[46] = -1.18, p = .243; boys with CAH v. unaffected boys: t[40] = 1.01, p = .317).

Figure 2: Interaction effect for GC.



A two-way ANOVA was conducted with FP acting as the dependent variable and with sex and condition acting as the fixed factors. A significant main effect of sex was observed ($F[1, 96] = 11.819$, $p = .001$, $\eta_p^2 = .113$) but the main effect of condition ($F[1, 96] = .907$, $p = .343$, $\eta_p^2 = .01$) and the interaction effects ($F[2, 95] = .79$, $p = .376$, $\eta_p^2 = .008$; see Figure 3) were not significant. These data suggest that boys felt more pressure for gender conformity than girls did, and that girls with and without CAH did not differ. Yunger, Carver, and Perry (2004) suggested that the source of pressure for gender conformity in children with Gender Identity Disorder might play a significant, varying role than in children from a normative population. They hypothesised that more pressure would originate from the external rather than the internal for this population. As such, all FP analyses were repeated examining between group differences in scores on the three FP source subscales: FP from parent, self, and peer (not shown). These analyses revealed no significant differences to those described above. Therefore, the total FP averages are used for the purposes of this examination.

Figure 3: Interaction effect for FP.

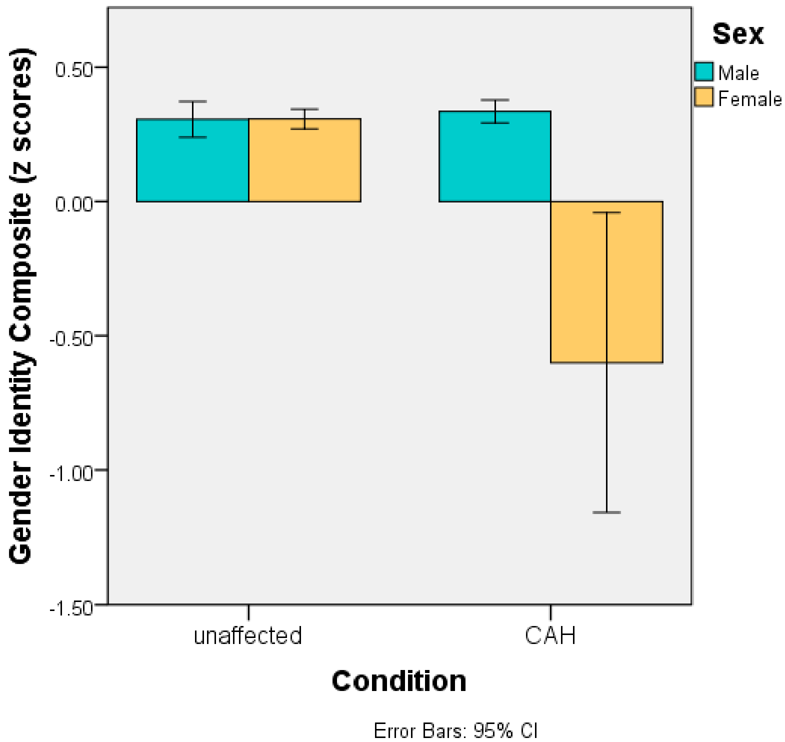


A two-way ANOVA was conducted with GI acting as the dependent variable and with sex and condition acting as the fixed factors. Significant main effects of sex (*F*[1, 96] = 9.90, *p* = .002, η_p^2 = .096) and condition (*F*[1, 96] = 8.73, *p* = .004, η_p^2 = .086) were observed as was a significant interaction effect (*F*[2, 95] = 9.96, *p* = .002, η_p^2 = .097; see Figure 4). Subsequent post-hoc pairwise t-tests revealed that the scores of the girls with CAH were significantly lower than the scores of all other groups (unaffected girls: *t*[25.22] = 3.34, *p* = .003; boys with CAH: *t*[25.29] = 3.45, *p* = .002; unaffected boys: *t*[25.69] = 3.32, *p* = .003) which did not differ significantly from one another (unaffected girls v. boys with CAH: *t*[50] = -1.04, *p* = .306; unaffected girls v. unaffected boys: *t*[29.52] = -.04, *p* = .968; boys with CAH v. unaffected boys: *t*[31.69] = -.786, *p* = .437).

Given the incongruence in variance of responses between groups for this variable (e.g. $SD_{(\text{unaffected girls})}$ = .10; $SD_{(\text{girls with CAH})}$ = 1.38), these analyses were repeated using the Kruskal-Wallis statistic, a non-parametric test robust in the face of data which lacks normal distribution. Significant group differences were detected (χ^2[3, 94] = 29.01, *p* < .01). Subsequent post-hoc pairwise Mann-Whitney U tests revealed that the scores of the girls with CAH were significantly lower than the scores of all other groups (unaffected girls: *p* < .001; boys with CAH: *p* < .001;

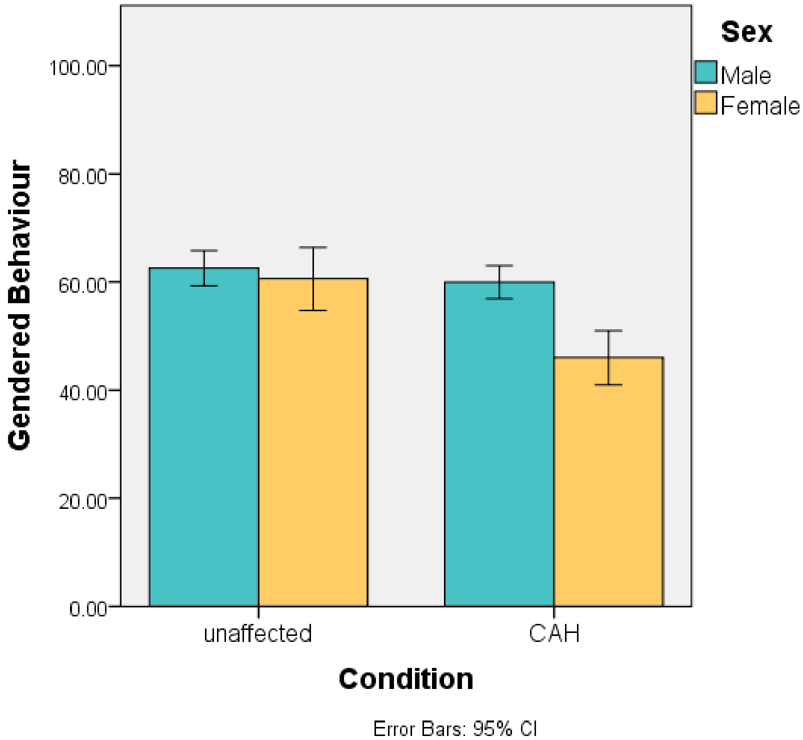
unaffected boys: $p < .001$) which did not differ significantly from one another (unaffected girls v. boys with CAH, $p = .170$; unaffected girls v. unaffected boys, $p = .602$; boys with CAH v. unaffected boys, $p = .662$).

Figure 4: Interaction effect for Gender Identity Composite.



A two-way ANOVA was conducted with GB acting as the dependent variable and with sex and condition acting as the fixed factors. Significant main effects of sex (F[1, 96] = 11.837, p = .001, $\eta_p^2 = .115$) and condition (F[1, 96] = 13.804, $p < .001$, $\eta_p^2 = .132$) were observed as was a significant interaction effect (F[2, 95] = 6.731, $p = .011$, $\eta_p^2 = .069$; see Figure 4). Subsequent post-hoc pairwise t-tests revealed that the scores of the girls with CAH were significantly lower than the scores of all other groups (unaffected girls: t[51] = -3.91, $p < .001$; CAH boys: t[40.72] = -4.91, $p < .001$; unaffected boys: t[40.26] = -5.75, $p < .000$) which did not differ significantly from one another (unaffected girls v. boys with CAH: t[38.81] = -.20, $p = .843$; unaffected girls v. unaffected boys: t[38.91] = -.61, $p = .547$; boys with CAH v. unaffected boys: t[40] = -1.20, p = .236).

Figure 5: Interaction effect for Gendered Behaviour.



An examination of the SDIGID, a measure designed to reflect the diagnostic criteria for GID (APA, 2000) revealed that the girls with CAH within our sample demonstrate a frequency of conforming to the criteria of GID diagnosis which was significantly higher than all other groups (χ^2[4, *N* = 90] = 4.38, *p* = .036; see Table 4). Based upon approximated frequency rates for adult females with GID in the general population (0.002-0.003%; APA 2013), this is an odds ratio of 7281.14. It is important to note that while this measure may reflect the criteria necessary for a GID diagnosis, it is not a diagnostic tool. For example, no measure of the extent to which gender variance caused distress to the participant was ascertained. Further, diagnoses usually occurs over multiple clinical visits. As such, what is indicated here is only a prediction of what diagnoses may have been received under the correct clinical circumstances and with the presentation of distress due to gender variance. That being said, while this method of diagnoses may not mimic perfectly that employed, it is nonetheless noteworthy that this comparison reveals a frequency of GID higher than what would be expected in the general population (Zucker & Lawrence, 2009).

Table 4: Numbers and Percentages of Children Meeting the DSM-IV-TR Criteria for Gender Identity Disorder (GID).

	Girls				Boys			
	CAH		Control		CAH		Control	
	N	%	N	%	N	%	N	%
Met DSM-IV-TR criteria for GID[†]	4/39*	15.4	0/38	0	0/36	0	0/31	0

Note: DSM-IV-TR criteria were met by endorsing "frequently" or "occasionally" for at least 4 of the following: (1) Wishes to be the other sex; (2) preference for cross-dressing; (3) preference for cross-sex role play; (4) preference for cross-sex pastimes; and (5) preference for other sex playmates. [†]All participants met criterion (1).
*$p < .05$ compared to each of the other 3 groups.

The overall, per sex, and per group correlations among each of the measures are displayed below (see Table 5). Several features of these correlations are worth note. First, while the overall correlations appear to indicate that all measures correlate quite strongly with one another, this is less true for felt pressure for gender conformity suggesting that there is something unique or different about this particular measure compared to the other measures being observed. For example, felt pressure for gender conformity only correlated significantly with gender typicality and gender identity scores and only for unaffected males (GT: $r[97] = .61$, $p < .01$; GT: $r[97] = -.56$, $p < .05$). This may suggest that something about having CAH may protect males from the relationships between felt pressure for gender conformity and feelings of gender typicality and gender ideations.

Second, it appears as though the between-measure correlations are driven by particular sex and group correlations. For example, the correlations between gender typicality and gendered behaviour were stronger for girls than boys ($p < .001$). This sex difference appears to be driven by the scores of girls with CAH. For girls with CAH, gender typicality correlated strongly with gendered behaviour ($r[97] = .69$, $p < .001$) and, to a lesser degree, with gender contentedness ($r[97] = .42$, $p < .05$). For the unaffected boys, gender typicality correlated significantly with all other measures except gendered behaviour (GC: $r[97] = .61$, $p < .01$; FP: $r[97] = .61$, $p < .01$; GI: $r[97] = .56$, $p < .05$). These group differences in the correlation between gender typicality and gendered behaviour suggests that, for girls with CAH, a unique association between these two factors exists. For unaffected boys, it appears that it is not the relationship between gender typicality and gendered behaviour that is most significant but rather the relationships with gender contentedness, felt pressure for gender conformity, and gender identity.

With regard to the main dependent variable, gender identity, it is worth noting that per sex correlations revealed that all measures correlated significantly with gender identity for females (GT: $r[97] = .40$, $p < .01$; GC: $r[97] = .51$, $p < .001$; GB: $r[97] = .51$, $p < .001$) except for felt pressure for gender conformity. Only gender typicality and felt pressure for gender conformity correlated significantly with gender identity for males (GT: $r[97] = -.39$, $p < .01$; FP: $r[97] = -.47$, $p < .01$). Per sex differences were found between the correlations of gender identity and gender contentedness ($p = .029$), felt pressure for gender conformity ($p = .038$) and gendered behaviour ($p = .018$). Per group correlations revealed that gender identity correlated significantly with gender contentedness and gendered behaviour for girls with CAH (GC: $r[97] = .42$, $p < .05$; GB: $r[97] = .69$, $p < .001$) while none of the examined factors correlated significantly with gender identity for boys with CAH. The only significant difference between the correlations of males and females with CAH was between the gender identity and gender contentedness variables ($p = .021$). No between group differences were observed between unaffected girls and girls with CAH nor between unaffected males and females. This suggests that for those with CAH, the relationship between gender identity and gender contentedness appears unique to girls.

Table 5: Correlations among Gender Measures Overall, Per Sex, and Per Group.

	Overall				
	GT	**GC**	**FP**	**GB**	**GI**
GT	--				
GC	.52**	--			
FP	.42***	.16	--		
GB	.55***	.38***	.25*	--	
GI	.44***	.53***	.17	.52***	--
	Per Sex				
GT	--	.46**	.43**	-.03	.39**
GC	.44***	--	.11	.27	.10
FP	.24	.05	--	-.07	.47**
GB	.60***	.31*	.29*	--	.06
GI	.40**	.51***	.07	.51***	--
	Unaffected				
GT	--	.61**	.61**	.07	.56*
GC	.21	--	.18	.39	.26
FP	.23	.02	--	-.20	.63**
GB	.31	-.12	.35	--	-.20
GI	.43*	.07	.12	.30	--
	CAH				

GT	--	.21	.06	-.08	.11
GC	.42*	--	-.08	.24	-.19
FP	.33	.08	--	.19	.12
GB	.69***	.39*	.33	--	.39
GI	.32	.47*	.13	.59**	--

* *p* < .05; ** *p* < .01; *** *p* < .001
Note: Overall correlations include all groups; per sex and per group correlations for boys are above the diagonal; per sex and per group correlations for girls are below the diagonal. GT – Gender typicality; GC – Gender contentedness; FP – Felt pressure for gender conformity; [†]GI - Gender identity composite – z-scores assessed; GB – gendered behaviour as per sex-typed CAI scores

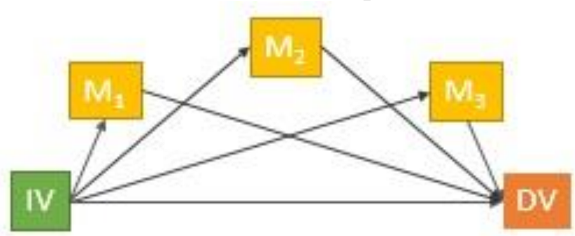
Due to weak statistical power from the sample size together with what would be a large number of intercorrelated predictor variables, multiple regression analyses were not suitable for these data. As such, alternative analysis types were sought out. Bootstrapping mediation using the program PROCESS, a non-parametric analysis more suitable for data that violates the assumption of normality and of a smaller sample size, was therefore chosen in order to examine the mediational/moderational roles of the relevant gender identity-related factors on the influence of prenatal androgen exposure typicality on gender ideations (see Preacher & Hayes, 2008 or Hayes, 2013 for a review of bootstrapping mediation/moderation). In brief, bootstrapping is a resampling method which treats the original sample as a miniature representation of the population. In mediation, an empirically derived representation of the sampling distribution of the indirect effect is generated upon which confidence intervals are based (Hayes, 2013, p. 106). As such, one can feel more assured making causal claims upon bootstrapping confidence intervals for this type of sample compared to alternatives (Preacher & Hayes, 2008; Hayes, 2013, p. 17, p.173).

Before describing the models that will be used in this analysis, it is important to distinguish between mediation and moderation (Hayes, 2013). Mediators are intervening variables to the relationship between two factors. In other words, *X* (the independent variable) is related to *Y* (the dependent variable) indirectly via a mediator; variation in *X* causes variation in *M* (the mediator[s]), which in turn causes variation in *Y*. This is different from moderation which occurs when the size or sign of the relationship between *X* and *Y* *depends* upon another variable, the moderator. Put another way, "whereas answering questions about *when* and *for whom* are the domain of moderation, questions that ask about *how* pertain to mediation…" (Hayes, 2013, p. 86). PROCESS is a free macro which can be used in combination of SAS or IBM SPSS in order to conduct these types of analyses in which a number of mediation and moderation formats have been designed to suit the needs of researchers and the various forms of hypotheses that may warrant testing (there are currently 76 possible models, see http://www.afhayes.com/public/templates.pdf for a complete list). The models used in subsequent analyses and outlined in more detail below were chosen to fit *a priori* hypotheses regarding the roles each gender identity-related factor might play in the relationship between prenatal androgen environment typicality and gender ideations. Because there were no group differences for felt pressure for gender conformity, it was not used in subsequent analyses.

The Model 4 mediation format allows researchers to determine whether multiple variables function as mediators simultaneously. Model 4 was therefore chosen in order to examine whether the remaining gender identity-related factors (e.g. gender typicality and gender contentedness) and gendered behaviour significantly mediated the relationship between prenatal androgen exposure typicality (girls with CAH v. all others) and gender ideations (as per the gender identity composite; n = 95; see Figure 6.1 for the model template). Results are displayed in Figure 6.2. The model was significant ($F[4, 90] = 16.99$, $p < .001$), and accounted for 43% of the variance. The direct effect of prenatal androgen exposure typicality on gender ideations trended toward significance ($t[94] = 1.93$, $p = .056$ suggesting that the relationship between X and Y is accounted for by the mediators being examined in this analysis. Gendered behaviour was a significant mediator as indicated by the significant indirect effect (Confidence Interval [95%] = .104, .573) while gender typicality (Confidence Interval [95%] = -.251, .253) and gender contentedness (Confidence Interval [95%] = -.079, .888) were not when examining these three variables as mediators simultaneously. These data support previous findings regarding the influence of prenatal androgens on gendered behaviour. It is of note, however, that when gendered behaviour is examined as a mediator on its own, the direct effect of prenatal androgen exposure typicality on gender ideations is significant ($t[94] = 3.12$, $p = .002$).

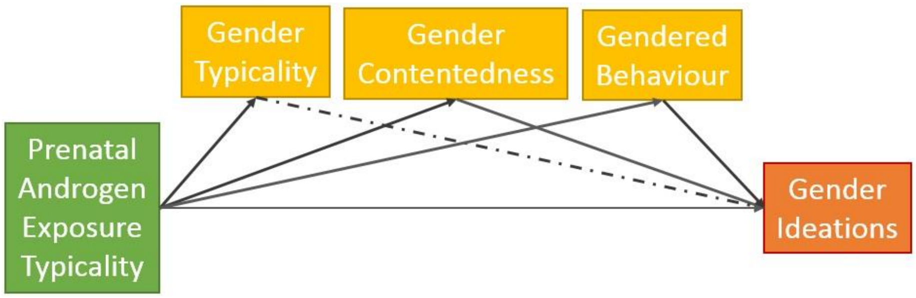
Figure 6: Multiple Mediation using Bootstrapping

6.1: Model Template



IV – independent variable, DV – dependent variable,
M_n – mediator(s)

6.2: Model Results



Model: *F* (4, 90) = 16.99, *p* < .001
Direct effect of IV on DV, *t* (94) = 1.93, *p* = .056
Indirect effects of the Mediators:
GT, *CI* (-.251, .253)
GC, *CI* (-.079, .888)
GB, *CI* (.104, .573)

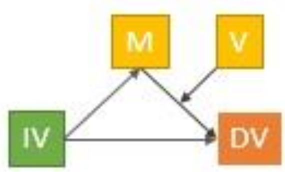
Note: Dark solid line, *p* < .001;
Light solid line, *p* = ns but is trending toward significant; Broken line, *p* = *ns*;
Bold indicates a significant Confidence Interval (95%).
Number of bootstrap resamples: 20,000

When the direct effect of X on Y is significant when examining the three variables as mediators simultaneously and not significant when examining one variable as a mediator on its own, it suggests that the other variables being examined in the first case may play a role in the relationship between X and Y, though this role is not as a mediator. In other words, while we can be reasonably assured that gendered behaviour significantly mediates the relationship between prenatal androgen exposure typicality and gender ideations, the role of gender typicality and gender contentedness in the relationship is less clear. Previous research on the development of gender suggest that, at least to some degree, interest in and expression of gendered behaviour likely occurs before more advanced gender self-cognitions like gender typicality and gender contentedness arise (Martin, Ruble, & Szykrybalo, 2002; Martin & Ruble, 2004). Therefore, as an extension of the above analyses and motivated by previous literature, it was hypothesised that gender typicality and gender contentedness might play moderating roles in the mediation of prenatal androgen exposure typicality on gender ideations by gendered behaviour. This hypothesis was tested. Given that a sense of gender typicality is necessary for the development of a self-appraisal of ones gender as satisfactory or not (e.g. gender contentedness), gender typicality was entered as a first-level moderator in the second stage of the mediation while gender contentedness was entered as a second-level moderator.

The Model 14 moderated mediation format was chosen to examine whether the mediation of the influence of prenatal androgen exposure typicality on gender ideations by gendered behaviour depended upon levels of gender typicality. Prenatal androgen exposure typicality (girls with CAH v. all others) was entered as the independent variable, gender ideations (as per the gender identity composite) was entered as the dependent variable, gendered behaviour was entered as the mediator, and gender typicality was entered as the second-stage moderator (n = 95; see Figure 7.1 for the model template). Results are displayed in Figure 7.2. The model was significant ($F[4, 90] = 15.67$, $p < .001$), and accounted for 41% of the variance. The direct effect of prenatal androgen exposure typicality on gender ideations was significant ($t[94] = 2.03$, $p = .046$). The moderated mediation of prenatal androgen exposure typicality on gender ideations by gendered behaviour was significant at low (2.18; Confidence Intervals [95%] = .151, .906) and medium (2.95; Confidence Intervals [95%] = .050, .498) levels of gender typicality.

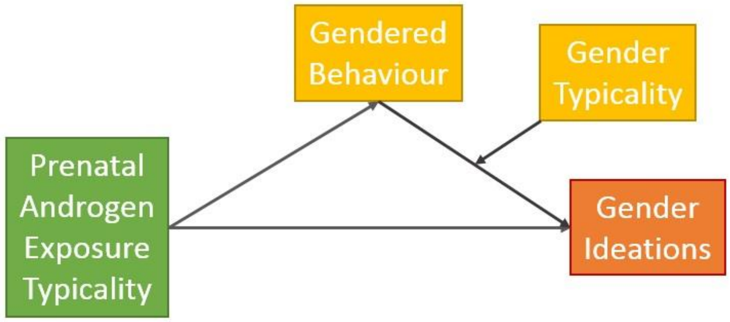
Figure 7: Moderated Mediation using Bootstrapping

7.1: Model Template



IV – independent variable
DV – dependent variable
M – mediator
V – second-stage moderator

7.2: Model Results



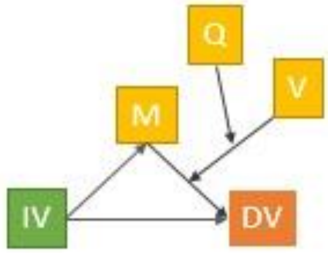
Model: *F* (4, 90) = 15.67, *p* < .001
Direct effect of IV on DV, *t* (94) = 2.03, *p* = .046
Moderated Mediation Effects:
GT = 2.18, *CI* (.151, .906)
GT = 2.95, *CI* (.050, .498)
GT = 3.72, *CI* (-.287, .187)

Note: Dark solid line, *p* < .001;
Bold indicates a significant Confidence Interval (95%).
Number of bootstrap resamples: 20,000

Model 18 moderated mediation format was then chosen in order to examine whether the moderated mediation of prenatal androgen exposure typicality on gender ideations by gendered behaviour depending upon levels of gender typicality also depended upon levels of gender contentedness. Prenatal androgen exposure typicality (girls with CAH v. all others) was entered as the independent variable, gender ideations (as per the gender identity composite) was entered as the dependent variable, gendered behaviour was entered as the mediator, gender typicality was entered as the first-level moderator, and gender contentedness was entered as the second-level moderator (n = 95; see Figure 8.1 for the model template). Results are displayed in Figure 8.2. The model was significant (F[8, 86] = 14.34, *p* < .001), and accounted for 57% of the variance. The direct effect of prenatal androgen exposure typicality on gender ideations was significant (*t*[94] = 2.31, *p* = .023) and the moderated, moderated mediation of gendered behaviour by gender typicality on the effect of prenatal androgen exposure typicality on gender ideations was significant at low levels of gender typicality (2.18) and at low (3.14) and medium (3.71) levels of gender contentedness (see Figure 8.2 for Confidence Intervals [95%]).

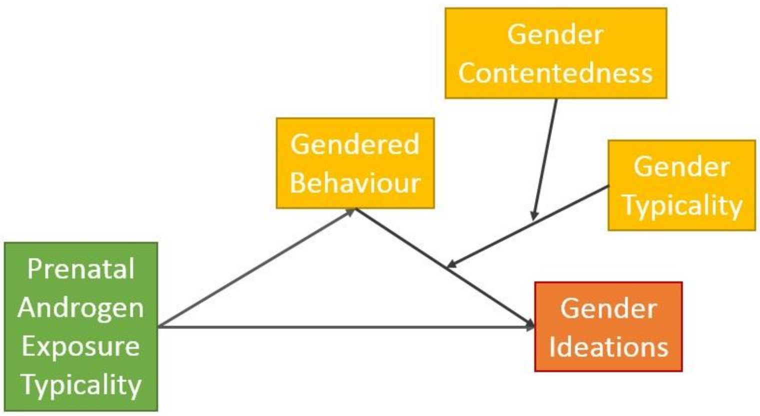
Figure 8: Moderated Moderated Mediation using Bootstrapping

8.1: Model Template



IV – independent variable
DV – dependent variable
M –mediator
V – first-level, second-stage moderator
Q – second-level, second stage moderator

8.2: Model Results



Model: F (8, 86) = 14.34, $p < .001$
Direct effect of IV on DV, t (94) = 2.30, $p = .023$
Moderated Mediation Effects:
GT = 2.18, GC = 3.14; *CI* (.192, 1.954)
GT = 2.18, GC = 3.71; *CI* (.033, .640)
GT = 2.18, GC = 4.00; *CI* (-.241, .450)
GT = 2.95, GC = 3.14; *CI* (-.136, 1.498)
GT = 2.95, GC = 3.71; *CI* (-.020, .501)
GT = 2.95, GC = 4.00; *CI* (-.224, .193)
GT = 3.72, GC = 3.14; *CI* (-.722, 1.500)
GT = 3.72, GC = 3.71; *CI* (-.293, .419)
GT = 3.72, GC = 4.00; *CI* (-.424, .224)

Note: Dark solid line, $p < .001$;
Bold indicates a significant Confidence Interval (95%).
Number of bootstrap resamples: 20,000

Discussion

This study is one of the first to evaluate gender identity as separate from gendered behaviour in a sample of children with and without CAH. It is also the first study to examine the influence of self-perceived gender typicality, gender contentedness, and felt pressure for gender conformity, on gender identity in the same population. Given the theoretical milieu of gender identity research, importance was placed on obtaining data from a large, age-specific sample, while using appropriate controls and previously standardized, self-report measures and attempting to minimise socially-desirable responding on measures assessing multiple domains of gender. As such, results presented here deal with a number of the limitations of previous examinations on gender identity in individuals with CAH. There are a number of observations that can be made based on the presented analyses.

Gendered behaviour and gender identity group differences. Past research has revealed inconsistency regarding whether girls and women with CAH present reduced female gender identity (or increased cross-sex ideations). Our findings replicate the bulk of research on the topic providing support for the hypothesis that girls with CAH do, indeed, present reduced female gender identity. Using the SDIGID, a measure reflecting the gender ideational and behavioural components of the DSM-IV-TR (2000) criteria for GID, prevalence rates which appear higher than those of control groups and higher than those expected in the general population further support this conclusion. These data also replicate previous findings whereby girls with heightened prenatal androgen exposure due to CAH appear to have masculinised/defeminised behaviour. Replication of previous findings conducted in a way that attempts to account for the limitations of previous analyses, such as the data presented here, provides increased assurance that the conclusions drawn are valid and reliable. These findings emphasise the need for practitioners dealing with the gender assignment of 46,XX females with CAH to consider the chance of later gender variance and to take appropriate precautions and actions including monitoring and providing support for cross-sex ideations and accompanying distress should they arise.

Gender typicality and gender contentedness group differences. In this sample, girls with heightened prenatal androgen exposure due to CAH exhibit decreased gender contentedness and gender typicality. Given that this is the first attempt to examine these particular constructs in a sample of children with CAH, these findings are novel and suggest that the heightened prenatal androgen exposure experienced by females with CAH might play a role in the development of self-perceived feelings of typicality with one's gender and the resulting contentment associated with this. The evidence that girls with CAH display a significantly decreased sense of self-perceived gender typicality when comparing themselves to other girls and a significantly decreased sense of contentment with being female confirms the need for researchers and practitioners to pay particular attention to the development of these feelings and the role they might play in later gender identity in girls with CAH. Future research would benefit from an analysis of factors which play a role in the development of feelings of gender typicality and gender contentedness.

Felt pressure for gender conformity sex differences. That boys (both with and without CAH) felt significantly more pressure for gender conformity than girls regardless of CAH status suggests that this particular gender identity-related factor may not be significantly associated with heightened prenatal androgen exposure but, rather, is a phenomenon unique to the male gender role. This observation is in line with other research suggesting that males feel more pressure to be sex-typed than females (Huston, 1983; Ruble, Martin, & Berenbaum, 1998) due to more rigid gender roles. The hypothesis that girls with CAH would be shifted in a male-typical direction for felt pressure for gender conformity was not supported. While this finding was not predicted, it does lend support to unpublished observations that girls with CAH appear to be resilient against feeling pressure for gender conformity. Pasterski et al. (2005) found that despite increased parental encouragement of gendered behaviour, girls with CAH persisted with the expression of gender nonconformity. The inability to detect or self-perceive pressure for gender conformity may be one mechanism of resilience for girls with CAH. In other words, an inability to detect pressure for gender conformity may be one factor which leads to a lack of response to encouragement of gendered behaviour. Because pressure for gender conformity is meant to cause children to avoid cross-sex activities and traits, lacking the male-typical shift in this gender identity-related factor suggests that these girls, despite displaying increased male-typical shifts in gendered behaviour, persist in gender nonconformity. However, it is also possible that the current measure for felt pressure for gender conformity was not sensitive to the particular pressures that may be felt by girls with CAH. For example, Yunger, Carver, & Perry (2004) suggested that more internal than external pressure for gender conformity might be felt by gender nonconforming children. Further research examining the role felt pressure for gender conformity might play in the development of gender identity in girls with CAH is needed. An examination into resilience against gender conformity in this population may also prove elucidating. Finally, future research would also benefit from an examination of the role felt pressure for gender conformity might play in male development and adjustment.

Egan and Perry (2001) originally hypothesised that felt pressure for gender conformity moderated the relationship between gender typicality and adjustment. This hypothesis was not supported by their data. They did, however, find that the effect of felt pressure for gender conformity on adjustment (e.g. social competence) was stronger for girls than for boys. While not presented here, our data did not support either hypothesis using gender ideations as a form of gender identity-related adjustment. As such, it appears as though felt pressure for gender conformity may play a stronger role in feelings of social competence than in the development of gender ideations or feelings of gender typicality. Felt pressure for gender conformity could be perceived as an infraction against one's feeling of general competence or compatibility as opposed to an infraction against one's gender competence in particular. For example, Yunger, Carver, and Perry (2004) found that children who felt strong pressure for gender conformity showed increased internalising problems and were less accepted by peers the following year. More research is needed to better understand what role felt pressure for gender conformity might play in individual well-being and adjustment and in gender variability.

Mediation and moderation by the gender identity-related factors. Fourth, analyses examining the relationship between the relevant gender identity-related factors (gender typicality and gender contentedness), gendered behaviour, and gender ideations found that gendered behaviour mediated the relationship between prenatal androgen exposure typicality and gender ideations. Data from this sample suggests that the experience and expression of gendered behaviour, which appears influenced by prenatal androgen exposure, has an impact on the development and expression of ones gender identity. This finding may not be surprising given that gendered behaviour appears to be one of the first components of a child's gender to develop and be expressed (e.g. eye gaze, toy, playmate, and activity preferences; Berenbaum & Hines, 1992; Connellan et al., 2000; Dittmann et al., 1990, Ehrhardt & Baker, 1974; Ehrhardt, Epstein, & Money, 1968; Hall et al., 2004; Hines, Brook, & Conway, 2004; Meyer-Bahlburg et al., 2004; Nordenstrom, Servin, Bohlin, Larsson, & Wedell, 2002; Pasterski et al., 2005, 2007, 2011). This observation also supports the use of gendered behaviour in the assessment of gender dysphoria given its role in the eventual expression of cross-sex ideation.

That gender typicality and gender contentedness were not significant mediators of the effect of prenatal androgen exposure typicality on gender ideations could reflect that these two gender identity-related factors do not play a role in the development of gender ideations to the degree that gendered behaviour does. If this is the case, it is possible that these gender identity-related factors may play more significant roles in the development of more general adjustment (e.g. self-worth). This conclusion is supported by the findings of Egan and Perry (2001). However, previous literature paired with the pattern of results of the current analysis motivated us to examine whether or not the mediation of prenatal androgen exposure typicality on gender identity by gendered behaviour might be moderated (as opposed to mediated) by gender typicality and gender contentedness. This hypothesis was support by these data. In other words, data from this sample suggest that the role gendered behaviour plays on the influence of prenatal androgen exposure typicality on gender ideations depends upon ones sense of gender typicality and the contentment derived from this self-perceived sense of typicality. Interestingly, of the per group average scores for gender typicality and gender contentedness, scores of girls with CAH most closely resembled values assigned automatically by PROCESS as “low” gender typicality and “low” gender contentedness. Attempts to decouple the impact of gendered behaviour and the impact of an individual’s self-appraisal of their gender on the development of their gender identity may be a promising avenue of exploration for researchers and practitioners studying typical gender development and for those looking to improve and tailor treatment of cross-sex ideation and the distress often associated with this (e.g. gender dysphoria), in children with GID and in girls with CAH.

Alternative Influences on Gender Identity in Females with CAH

Age and gender identity. Certainly, there are a number of other factors that could influence the development of gender identity that have not been examined here. As alluded to previously, age is an important factor related to the development of gender identity. Not only does gender identity development appear to be an ongoing process throughout childhood, puberty also appears to play an important role (Steensma, Biemond, de Boer, & Cohen-Kettenis, 2011). This may be particularly true for those experiencing gender dysphoria. Of children with GID/GD, only 15% appear to remain gender dysphoric through puberty and into adulthood (Steensma et al., 2013). Children with the most extreme presentation of gender variance and dysphoria appear to be the most likely to persist. Further, persistence rates of females with GID/GD appear to be higher than for males (Wallien & Cohen-Kettenis, 2008). In their analysis, Steensma and colleagues (2011) point to three factors as possible contributors to the desistence and persistence of GID: 1) physical puberty (e.g. hormone fluctuations, physiological development); 2) the changing environment and the explicit treatment as one's natal sex within this new environment (e.g. high school); and 3) the discovery of sexuality. That there were no significant effects of age on the constructs being examined in the current study is an important feature of this analysis because it suggests that there may be, at least to some degree, congruity among the children in our groups for the constructs being examined as a function of age and may reflect the children not yet having entered puberty. As such, the gender variance detected hasn't yet been influenced by the above contributors and can be thought of as components of the expression of childhood gender. Future research should include longitudinal examinations following children with CAH through puberty and into adulthood. Longitudinal analyses in this population would provide researchers with the ability to better understand the role the gender identity-related factors might play at various phases of gender development including the ability to observe changes in the various components associated with gender identity over time. Analyses following participants through stages important for gender development would help establish rates of persistence and desistence for those with CAH as well as elucidate the role the contributors outlined by Steensma et al. (2011; puberty, changing environment, and sexuality) might play in later gender identity.

CAH as a life-long condition. While the relationship between age and gender variance has received research attention, the role having a chronic condition might play on gender identity development has not. CAH is a condition that necessitates life-long management and is typically treated with the on-going administration of cortisol (commonly hydrocortisol and/or other forms of glucocorticoids [GCs] and mineralocorticoids [MCs] depending on the severity of the condition; Newns, 1974; Speiser et al., 2010). Due to the short half-lives of the drugs used, administration occurs 2-3 times daily. For the first week after diagnosis, GCs/MCs must be administered in a high dose to suppress adrenocorticotropic hormone (ACTH) production, a hormone responsible for the stimulation of the production of androgens. Dosage is later dropped to a maintainable level. Later on, it is often necessary for the administration of GCs/MCs to be temporarily increased and overseen in response to heightened stress and physiological emergency. Dosage management and monitoring is done regularly (every 3 months during infancy; every 6 months during childhood) and consists of measuring height, weight, bone age, and hormone levels (17-OHP, androstenedione, and testosterone). As a check for treatment efficacy, high 17- OHP levels, adrenal crisis and evidence of hyperandrogenism (e.g. loss of growth potential, precocious puberty, increased bone age) suggest *undertreatment* while low 17-OHP levels and hypercortisolism (e.g. suppressed linear growth, bone age retardation, Cushing's syndrome) suggest *overtreatment*. Therefore, a balance to avoid the negative consequences of CAH and the negative consequences of its treatment is sought. As a patient's hormone environment typically stabilises into adulthood, endocrine management commonly wanes while medication use continues (Newns, 1974; Speiser et al., 2010).

It is not surprising, then, that the above treatment and management can be disruptive to the lives of those with the condition. The impact of having CAH and the subsequent impact treatment of this chronic condition are additional factors that may influence gender identity development for this population. In particular, the GCs/MCs that patients with CAH are treated with could contribute to the development of later gender variance and dysphoria. Such treatment is often associated with Cushoid-like symptoms (e.g. increased adiposity, short stature, elevated anxiety), characteristics that can impact subsequent psychological well-being (e.g. body image). In fact, children and adolescents with CAH have higher body mass index (BMI) than matched controls (Cornean, Hindmarsh, & Brook, 1998; Völkl, Simm, Beier, & Dörr, 2006; Völkl et al., 2009). Further, heightened BMI (due to shortened stature and increased abdominal adiposity) in those with CAH has been linked to cortisol administration (Bonfig, et al., 2009; Darmon et al., 2006; Knorr & Hinrichsen-de-Lienau, 1980). Finally, Kuhnle, Bullinger, and Schwarz (1995) found that CAH patients felt significantly less attractive, less pleasant in appearance, were shyer because of their appearance, and were significantly more likely to indicate wanting a new body than controls. This same study also found that CAH women were more negative in their self-image due to significantly lowered self-confidence, sociability, and social acceptance compared to controls. As such, disrupted psychological well-being and variable body morphology could play a role in the development of one's gender identity and could lead those with CAH to feel less congruent with other members of their gender. For example, variable body morphology could lead to decreased body image resulting in less engagement in activities that could promote feelings of gender congruity. The treatment of CAH and the ramifications of its overtreatment stand as largely un-researched but potentially important influences upon the development of gender and gender identity.

In addition to the effects of the treatment of CAH, having a chronic condition also means regular management of the condition itself. Frequent medical check-ups and endocrine testing, dose monitoring, dietary and weight management, and the need for daily and emergency medication throughout the lifespan are all features of having CAH. Each of these influences to daily life are also in addition to the possibility of the impact of surgery (discussed in detail below). The nature of these tasks is disruptive, necessitates careful planning and scheduling, and can impede engagement in other activities (Gallant, 2003). As such, self-management of a chronic condition could pose an additional influence to the development of gender and gender identity. For example, a child might be denied permission to engage in particular activities due to the risk of heightened stress or injury. Time spent in hospital is time that might otherwise be devoted to activities that could be identity enforcing including participation in clubs and sports or time spent with peers. It is not uncommon for females with CAH to recall the substitution of regular childhood summer activities for a stay at hospital. In sum, an important avenue for future research could include an examination of the role the impact of having a chronic condition and its management has on gender and gender identity development.

CAH and genital ambiguity. In addition to the effects of having a chronic condition and its treatment and management, CAH also exerts particular influence on the genital physiology of females with the condition. Excess prenatal androgen exposure masculinises the external genitalia of 46,XX infants with CAH to varying degrees depending on severity of the condition (Dittmann et al., 1990; Pang et al., 1985). For 46,XX patients with low vaginal confluence (communication between the vagina and urinary tract; Hasan, Tasmin, Salam, & Talukder, 2011), genital surgery during infancy may be necessary. Labial fusion or urinary tract malformation can risk the health of the infant and, as such, can necessitate surgical interventions including vaginoplasty, perineal reconstruction, and/or clitoroplasty (Speiser et al. 2010). Such intervention can, in turn, increase risk of repeat surgery, introduce scar tissue, and decrease plasticity, all of which may compromise later sexual function, sensitivity, and satisfaction (Callens et al., 2012; Crouch, Liao, Woodhouse, Conway, & Creighton, 2008; Minto, Liao, Woodhouse, Ransley, & Creighton, 2003; Van der Zwan et al., 2013; Wisniewski, Migeon, Malouf, & Gearhart, 2004). It is currently unknown what impairment may result if non-life threatening malformations are left un-manipulated.

Ambiguous genitalia propose a specific challenge for medical professionals and researchers of gender identity development. Given the influence of prenatal androgen exposure on the external genitalia and possibility of subsequent genital surgery in 46,XX patients with CAH, the role of genital appearance and function on gender identity development has been examined. In the past, it was hypothesised that growing up with genitals that were incongruent to ones assigned sex would lead to gender confusion or dysphoria. The theory that assigned gender and external genitalia need be congruent for the development of a healthy gender identity was proposed by Dr. John Money and colleagues (Money, 1975; Money & Ehrhardt, 1972) and was supported by a number of practitioners managing conditions resulting in ambiguous genitalia. This view, together with the possibility that the genital ambiguity could negatively impact the physical health left genital feminising surgery as common practice for infant girls with CAH (see Dreger, 1998 for review), even in cases of mild to moderate masculinisation. Follow-up research and public attention put this practice under scrutiny and evidence eventually demonstrated that the development of gender identity is influenced by more factors than congruity between genital appearance and sex-of-rearing alone. Decades later, the decision to perform surgery on infants born with genital ambiguity is now made by a team of practitioners based on a plethora of factors including degree of physical functioning, the possibility of future functioning and reproductive ability, severity of masculinisation, parent preference, and possibility of treatment adherence (Hughes et al., 2006). As such, genital appearance and function (even in the context of those whose sex of rearing and genital appearance are congruent) could play a role in later gender identity development for females with CAH. As described above, the development of one's sexuality occurring around puberty is hypothesised to play a role in the development of later gender identity. Genital appearance and function could play a particular role in the influence puberty and ones sexuality has on gender identity development. For example, surgery can adversely affect the experience of sexual pleasure (Boyle, Smith & Liao, 2005; Minto et al., 2003) and, depending on the type of surgery, can result in decreased sexual activity (Minto et al., 2003). Decreased sexual function and activity, especially during puberty when one's sexuality is developing and being explored coupled with the direct impact puberty appears to play in gender identity solidification, may influence the development of ones gender identity. In fact, women with CAH report less sexual and romantic activity (Zucker et al., 1996) and decreased sexual arousability (Zucker, Bradley, Oliver, Blake, Fleming, & Hood, 2004) than controls. Even for a group of adults with DSD's, making the decision to undergo surgery and the process of undergoing genital surgery presents a psychological dilemma (Boyle et al., 2005). Reviews such as these emphasise the wide-ranging impact decisions about surgery and the results of these surgeries could have on the lives of those born with ambiguous genitalia.

Limitations and Future Research

I have already alluded to the primary weakness of this study - the size of the sample. Obtaining a sample size which is sufficient for the conduction of the ideal types of statistical analyses for testing particular hypotheses is a common challenge when working with populations with rare conditions. While our sample was larger than most studies examining gender identity in individuals with CAH, a larger sample would decrease the likelihood of Type 1 error and increase the types of statistical tests that could be performed. While using the style of statistical test utilised in the current analysis (bootstrapping mediation/moderation) can facilitate and add robustness to analyses upon samples of this size, no statistical test can replace a sufficient sample size. Further, given that this examination set out to examine gender identity *development* in particular, adding a longitudinal component to this analysis would greatly support the conclusions being drawn. By observing this population and measuring the factors hypothesised to influence gender identity over time, we might begin to observe a more complete view of how factors like self-perceived gender typicality, gender contentedness, felt pressure for gender conformity, and gendered behaviour impact gender identity development. As such, the cross-sectional nature of these data stand as an additional weakness to this analysis.

An empirical consideration and examination of the impact of the proposed alternative influences of gender identity development would also facilitate an improved and more holistic understanding of how gender identity develops in individuals with CAH. As it stands, research on the topic suggests that age and the genital appearance and function of females with CAH may play a role in the development of their gender identities. As discussed, potential genital appearance and function variation such as that experienced by girls with CAH, together with subsequent genital self-image (or feelings regarding the appearance and functioning of one’s genitals), could influence sexual development during puberty, which might, in turn, influence ones gender identity. This may be especially true for children with GID/GD who often express a discomfort, disgust, or inappropriateness of their genitals (Zucker, 2010). The carrying of these evaluations through puberty could negatively impact sexual development and exploration. As such, the gender identity-related factors (gender typicality and gender contentedness), gendered behaviour, genital appearance and function, and age in children with CAH could play an interactional role on the development of their gender identities. As our understanding and use of statistical tests needed for examining these interactional processes advance, these types of examinations should be considered. Further, both the influence of CAH treatment and the impact of having a chronic condition warranting high levels of self-management stand as largely under-researched features of CAH which could influence gender and gender identity development and are likely important avenues for future research.

Implications and Conclusion

In sum, these findings have implications for the current theoretical understanding regarding gender identity development and emphasise that the expression of gendered behaviour plays an important role in this development. Moreover, it appears that the role of gendered behaviour on gender identity development may depend upon self-appraised feelings of typicality in one's gender and the contentment derived from this appraisal. For girls with CAH (a population suspected to display reduced female gender identity), these data provide further support for the role heightened prenatal androgen exposure plays on the expression of gendered behaviour, gender typicality, gender contentedness, and gender identity. Further, it appears that for girls with CAH, expressing cross-sex gendered behaviour, lacking feelings of gender typicality, and lacking contentment with one's gender play significant roles in the subsequent development of their gender identities (including those expressing cross-sex gender ideations). In this sample, it is not only the interest and expression of cross-sex gendered behaviour that influences cross-sex ideation in girls with heightened prenatal androgen exposure but also how one self-appraises (both with regard to the extent to which one feels typical and content with one's gender) this gendered behaviour. The moderational role of self-appraised gender typicality and gender contentedness highlights the importance of individual variability and assessment of ones gender and the role this might play in the subsequent experience of distress associated with gender variance. As such, attempts to improve and negate the impact of the distress associated with cross-sex ideation might be positively benefitted by providing early monitoring and support for feelings of low gender typicality and contentedness as an addition to treatments focused on the presentation of gendered behaviour. Research into factors that influence the development of these gender identity-related factors, such as internal and external gender-related stigma, may reveal promising avenues for improvement to early treatment of distress associated with cross-sex ideation.

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Appendix 1: Factor loadings for the *Structured Diagnostic Interview for Gender Identity Disorder* (SDIGID) and the *Gender Identity Questionnaire for Parents* (GIQ).

Measure	**Maintained Items**	**Factor Loadings***		
		1	**2**	**3**
SDIGID	Cross-dressing	.572	.354	.098
SDIGID	Role play	**.860**	.189	.081
SDIGID	Peer relations (same or other sex)	**.602**	.422	.102
SDIGID	Games and pastimes	**.778**	.164	.042
GIQ	Playmates (same or other sex)	**.644**	.252	.148
GIQ	Plays with Barbie	**.767**	.149	-.432
GIQ	Plays with make-up	**.764**	.206	-.342
GIQ	Imitates female characters	**.658**	.276	-.495
GIQ	Plays sports with boys (but not girls)	**.610**	.209	.009
GIQ	Plays same sex in playing house	**.699**	.220	.024
GIQ	Plays girl-type games (compared to boy-type)	**.704**	.176	-.176
GIQ	Plays boy-type games (compared to girl-type)	**.721**	.160	.085
GIQ	Dresses up in same or other sex clothes	**.815**	.295	.037
SDIGID	States that s/he is the other sex	.321	**.892**	.055
GIQ	States wish to be other sex	.276	**.829**	.088
GIQ	States that s/he is the other sex	.241	**.613**	-.068
Measures	**Discarded Items**			
GIQ	Imitates male characters	.424	.069	**.664**
GIQ	Plays sports with girls (but not boys)	-.371	.098	.241
GIQ	Plays with GI Joe	-.029	-.106	**.775**

*Only the strongest loadings for each item are presented in the table,
Notes: Items greater than .6 are bolded. Factor 2 items were used for the gender identity composite. Other items were used in analyses conducted for a different manuscript submitted for publication.

Appendix 2: Factor loadings for *Gender Typicality Scale.*

Items Used	Factor Loadings	
	1: Other Gender	**2 : Same Gender**
3. Some girls have the same interests that boys have, but…	**.750**	.226
7. Some girls sometimes play with boys' toys and do boys' activities, but…	**.759**	.227
8. Some girls feel their personality is different from most girls' personalities, but…	.129	**.790**
13. Some girls have the same feelings that boys have, but…	**.658**	.141
14. Some girls feel they are different from other girls, but…	.182	**.689**
16. Some girls feel that their personality is different from boys' personalities, but…	**.853**	.261
19. Some girls never talk or act like a boy, but…	**.702**	.216
Items Discarded	**Factor Loadings**	
	1: Other Gender	**2 : Same Gender**
1. Some girls have the same interests that other girls have	.332	.398
5. Some girls don't talk or act like other girls, but…	.253	.419
10. Some girls feel they are very different from boys, but…	.587	.154
11. Some girls feel that the things they like to do in their spare time are similar to what most girls like to do in their spare time, but…	.432	.586
17. Some girls have the same feelings that other girls have, but…	.143	.528
20. Some girls like to play with the same toys that other girls do, but…	.515	.538
22. Some girls don't like the same activities that other girls like, but…	.586	.503

Note: Items greater than .6 are bolded, rotated factor matrix displayed here. Girl form.

Appendix 3: Factor loadings for *Gender Contentedness Scale*.

Items Used	Factor Loadings	
	1:	**2:**
2. Some girls are happy that they are a girl, but…	**.755**	.199
4. Some girls do not love being a girl, but…	**.610**	.246
15. Some girls are glad they'll grow up to be a woman, but…	**.749**	.249
21. Some girls wish they didn't have to be a girl all their life, but…	**.825**	.353
Items Discarded	**Factor Loadings**	
	1:	**2:**
6. Some girls daydream more about doing boys' activities, but…	.239	**.919**
9. Some girls like girls' hobbies and interests more than boys' hobbies and interests, but..	.238	**.790**
12. Some girls don't like being a girl, but…	.522	.164
18. When they watch a movie, some girls find the boy characters more interesting than the girl characters, but…	.237	.406

Note: Items greater than .6 are bolded, rotated factor matrix displayed here. Girl form.

Appendix 4: *Felt Pressure for Gender Conformity* items.

Items Used
1. My parents want me to be similar to other girls.
2. The girls I know would be upset if I wanted to play with boys' toys.
3. I don't think it's important for me to do the same activities that other girls do.*
4. I would be ashamed of myself if I heard myself talking like a boy.
5. The girls I know would think it was OK if I talked or acted like a boy.*
6. I think it's important for me to be good at the same activities that other girls are good at.
7. My parents would be upset if they saw me acting like a boy.
8. The girls I know don't care whether I act like other girls or not.*
9. I think it would be wrong for me to play with boys' toys or do boys' activities.
10. The girls I know think it's important that I have the same interests as other girls.
11. The girls I know wouldn't like it if I wanted to learn an activity that boys usually do.
12. My parents don't think it's important for me to act like other girls.
13. I would be upset with myself if I didn't act like other girls.
14. My parents wouldn't mind if I wanted to collect things that boys usually collect.
15. I wouldn't like myself if I heard myself talking or laughing like a boy.
16. My parents would be upset if I didn't act like other girls.
17. The girls I know wouldn't mind if I wanted to play a boys' sport.*
18. My parents would think it was OK if I wanted to play with boys' toys.
19. The girls I know would be upset if I didn't want to do the same activities that other girls do.
20. I would still like myself if I saw myself acting like a boy.*
21. My parents want me to do the same activities that other girls do.
22. I don't think it's important for me to be similar to other girls.
23. My parents wouldn't like it if I wanted to learn an activity that only boys do.
24. The girls I know don't think it's important for me to act like other girls.

Note: Girl form.