

Abstract

The present research used linked surveillance systems (British Paediatric Surveillance Unit; and the Child and Adolescent Psychiatry Surveillance System) over a 19-month period (1 November 2011 – 31 May 2013) to notify of young people (4-15.9 years) presenting to secondary care (paediatrics or child and adolescent mental health services) or specialist gender services with features of GD. A questionnaire about socio-demographic, mental health details, and GD features was completed. Presence of GD was then assessed by experts in the field using the criteria at the time (DSM-IV-TR). Incidence across the British Isles was 0.41-12.23 per 100,000. 230 confirmed cases of GD were noted; the majority were white (94%), aged ≥ 12 years (75.3%), and were assigned female at birth (57.8%). Assigned males presented most commonly in pre-adolescence (63.2%), and assigned females in adolescence (64.7%). Median age-of-onset of experiencing GD was 9.5 years (IQR 5-12), the majority reported long-standing features (2-5 years in 36.1%, ≥ 5 years in 26.5%). Only 82.5% attended mainstream school. Bullying was reported in 47.4%, previous self-harm in 35.2%, neurodiversity in 16%, and 51.5% had ≥ 1 mental health condition. These findings suggest GD is rare within this age group and that monitoring wellbeing and ensuring support for co-occurring difficulties is vital.

Keywords: Gender dysphoria, child, adolescent, mental health, transgender, gender diverse, gender distress

Introduction

It is not unusual for young children to express their gender in ways which may differ from societal expectations associated with assigned gender (Gray, Carter & Levitt, 2016). However, some children and adolescents experience profound distress around birth assigned gender and identified gender, or significant impairment in functioning. When this distress or impairment is evident over ≥ 6 months, the Diagnostic and Statistical Manual of Mental Disorders version 5 (DSM-5) indicates that gender dysphoria (GD) is present. The WPATH Standards of Care (version 7) and ICD-11 note that not all gender diverse individuals experience GD, and those that do may not experience it throughout their lives. Indeed, children who show gender diverse behaviour only do not fulfil the criteria for GD. There is no established single causation for GD. Research surrounding aetiology is inconclusive (e.g. evidence for a biomedical causal model remains limited; Drescher & Byne, 2012; Bao and Swaab 2011). Instead, GD is most likely experienced due to a combination of biological and sociocultural factors.

Prevalence

Existing epidemiological studies of GD are predominantly of adolescents ≥ 15 years and adults, typically involving those who have accessed specialist gender clinics. The findings vary widely across studies, reflecting different methodologies and differences between countries in treatment availability, social acceptability, diagnostic criteria, and treatment eligibility criteria (Aitken et al., 2015; Skordis et al., 2020). Prevalence estimates of identifying as transgender ranged from 1:2000 (approximately 0.05%) in the Netherlands and Belgium (Olyslager & Conway, 2007) to 1.2% in New Zealand (Clark et al., 2014). Social acceptability of attending gender services must also be taken into account (e.g. considering cultural factors affecting access; de Graaf et al., 2019). Nevertheless, the number of young

people referred to specialist gender clinics for GD appears to be increasing (e.g. de Graaf et al., 2018; Kaltiala-Heino, 2020). This may relate to changing help-seeking attitudes, reduced stigma, increased visibility, and raised public awareness (Skordis et al., 2020). International studies also show a shift in the sex ratio of adolescent referrals from favouring assigned males at birth to favouring assigned females at birth (Aitken et al., 2015; de Graaf et al., 2017; Kaltiala-Heino et al., 2020).

Puberty and Mental Health

In many children, GD will fade before, or early in, puberty (Ristori & Steensma, 2016; Kaltiala-Heino et al., 2018). However, in some these feelings intensify and body aversion can develop or increase as puberty progresses. Adolescence, with its changing social environment and onset of physical puberty, may contribute to continued experience of GD (Steensma et al., 2013). Suicidal ideation and self-harm are more prevalent in young people with GD as compared to cis-gender individuals, and may be related to discrimination and victimisation (de Graaf et al., 2020; Almeida et al., 2009). Consistent experience of minority stressors such as micro- and macro-aggressions, social rejection, stigma, transphobia and a lack of support structures have been indicated as likely contributors to mental health concerns among gender diverse individuals (Chodzen et al., 2019; Delozier et al., 2020). Internationally, clinic-referred children and young people experiencing GD show increased incidence of neurodiversity (e.g. autism spectrum conditions; (ASC) van der Miesen et al., 2018). More recent systematic reviews suggest prevalence rates of ASC in GD individuals, although higher than general population averages, may be similar to those seeking psychiatric care (Thrower et al., 2019).

Owing to variation in reportage of numbers, demographics and mental health difficulties experienced by young people with GD, due to varied methodologies and the criteria for inclusion employed, this research was deemed vital to generate estimates of (i) incidence, (ii) socio-demographics, (iii) mental health difficulties and (iv) management of young people accessing secondary care or specialist gender services in the British Isles. Such work is needed to inform continuing development of appropriate assessment, support, and referral pathways to specialist gender services.

Methods

Design:

This study was conducted over 19 months (1 November 2011 – 31 May 2013) using two linked surveillance systems operating across the British Isles (United Kingdom and Republic of Ireland).

The British Paediatric Surveillance Unit (BPSU) reporting system is well accepted for the study of 'rare childhood conditions' (Hall & Glickman, 1988). Consultant paediatricians throughout the British Isles are sent monthly notification cards listing health conditions currently under study. Notification cards are returned to BPSU administrators who inform the relevant research team when a clinician reports a case. A questionnaire is then sent to the reporting clinician for further details about the case. The Child and Adolescent Psychiatry Surveillance System (CAPSS) uses identical methodology to the BPSU, sending out a monthly report card to consultant child and adolescent psychiatrists.

Ethical consideration:

National Research Ethics System approval: London Bloomsbury REC (REC Ref: 11/LO/1512). National Information Governance Board (NIGB) approval was obtained (under Section 251 of the NHS Act 2006; ref: ECC 8-02(FT4)/2011) for notification without the need for consent from children or families.

Study population:

Children and young people aged 4.0-15.9 years presenting to secondary care (paediatrics or child and adolescent mental health services (CAMHS)) or specialist gender services in the British Isles were eligible for inclusion. Those with differences in sex development (DSD) conditions or major psychotic disorder were excluded.

Case definition:

The surveillance case definition used (**Figure 1**) was derived from the DSM-IV-TR criteria for GD, in use at the time of data collection. The criteria were modified to encourage over- rather than under-reporting to ensure more subtle cases were not overlooked. Clinicians were asked to notify all new cases meeting the surveillance case definition seen for the first time in the last month. Cases notified by specialist gender services (the UK Gender Identity Development Service (GIDS) or the Sandyford Gender Identity Service in Scotland) were captured via the CAPSS surveillance system.

Figure 1. Surveillance case definition criteria for case notification

BOTH the following criteria (1 and 2) should be fulfilled:

1. A strong cross-gender identification for ≥ 6 months

- (i) In children <12 years, this requires 2 or more of the following:
 - a) In boys, preference for cross-dressing or simulating female attire; in girls, insistence on wearing only stereotypical masculine clothing;
 - b) Strong preferences for cross-sex roles in make-believe play or fantasies of being the other sex;
 - c) Intense desire to participate in the stereotypical games and pastimes of the other sex;
 - d) Strong preference for playmates of the other sex.

- (ii) In adolescents ≥ 12 years, this requires 1 or more of the following:
 - a) Frequent passing as the other sex (adopts clothing, hairstyle of the other sex)
 - b) Desire to live and be treated as the other sex
 - c) Belief that their feelings and reactions are typical of the other sex

2. a) Distress or unhappiness with his/her biological sex

(e.g. Stated dislike of/aversion to or self-inflicted injury to their primary or secondary sexual characteristics, request for physical intervention to alter their physical sexual characteristics to those of the other sex)

OR

- b) Stated desire to be or belief that he/she is or should be the other sex**

Data collection:

Clinicians notifying potential cases completed and returned a questionnaire for each case including demographic details; GD features, their duration and age at onset; referral source; co-occurring mental health difficulties at presentation; social history including family make-up and schooling; and data on clinical management and health service use (see **Supplementary Material 1**). Reminders were sent out if questionnaires had not been returned. Follow-up data were obtained from the same clinicians at 1 and 2 years; this paper focuses on the initial baseline data of the recognised cohort.

Case validation and handling of duplicates:

Cases were validated against the diagnostic (DSM-IV-TR) case definition (**Supplementary Material 2**) by a panel with clinical expertise. Duplicates were identified using personal identifiers (NHS number, date of birth, first part of post code). For BPSU/CAPSS duplicates, the CAPSS date of diagnosis was used. For duplicates within the CAPSS system (e.g. cases notified by a psychiatrist and then by a specialist gender service), the earlier date of diagnosis was used. In a few cases with a diagnosis of ASC, evidence of GD was present despite not fulfilling all behavioural criteria. These cases were included on the basis of potential impact of ASC. Where there were discrepancies in GD features reported between services, specialist gender clinic reportage was accepted over other services, followed by psychiatrist, then paediatrician symptom reportage. For discrepancies in reported mental health difficulties, psychiatrist reportage was prioritised. Where data were provided by one reporting clinician and omitted or 'not known' by another (e.g. regarding mental health difficulties or social history), these data were included.

Analysis:

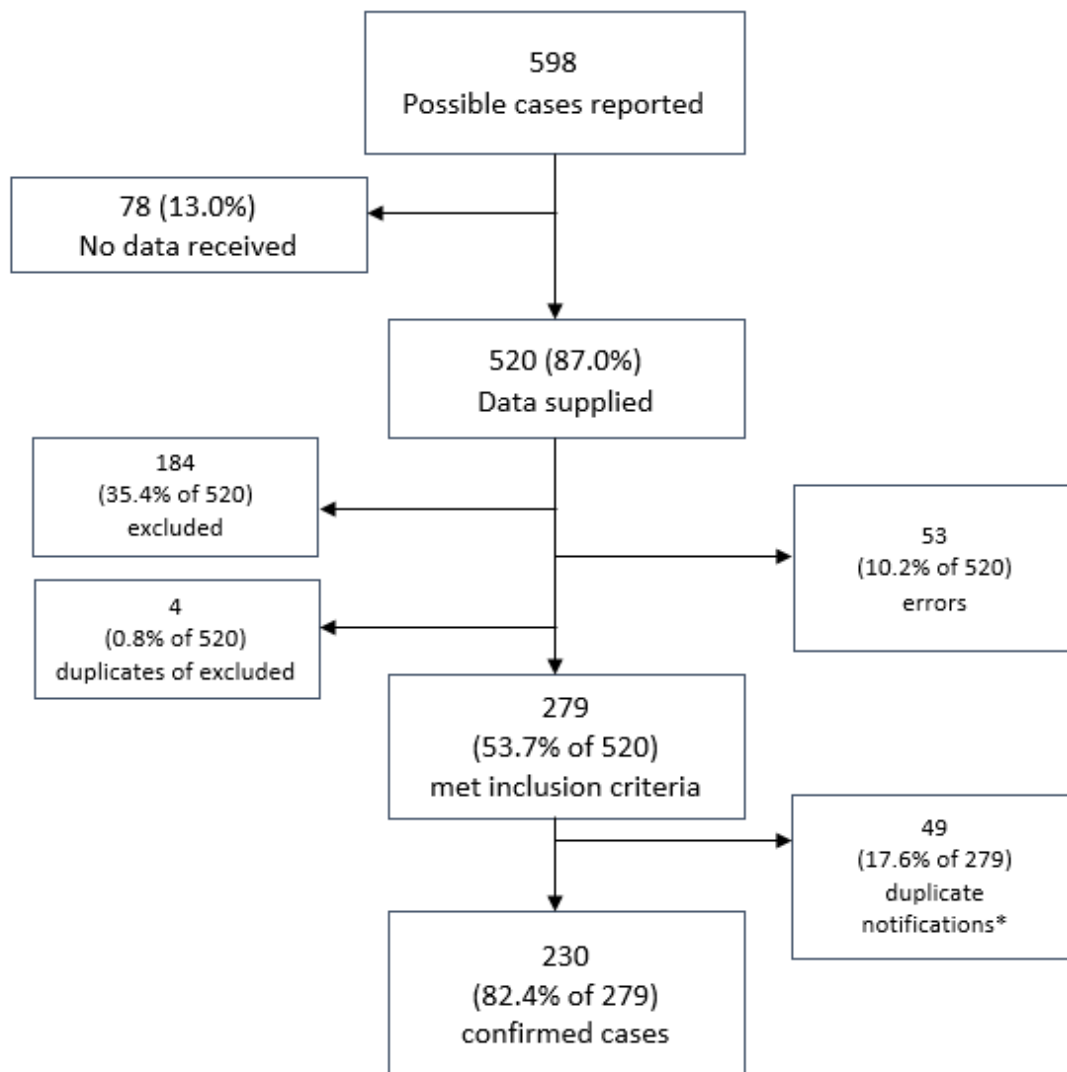
All analyses were descriptive, and used SPSS 27 (IBM analytics). Age at onset of features was categorized into the following age brackets: <5 years, 5 to 11 years, and ≥ 12 years. Duration of GD features was categorized into the following sub-groups: <2 years (6 – 23 months), 2 to 5 years and ≥ 5 years. Incidence was estimated by aggregating cases at a country level and calculating the incidence in a 12-month period per 100,000 population by country, and for the UK countries as a whole at that time.

Results

Participants

The notification card return rate over 19 months was 93% for the BPSU and 76% for CAPSS. Five hundred and ninety-eight cases were notified, 140 by the BPSU and 457 by CAPSS, with an additional case notified directly by their general practitioner (GP) to the BPSU. The flow of individuals from notification to case validation is shown in **Figure 2**. Potential cases were excluded if no response was obtained after multiple attempts to contact the notifying clinician or their team (n=78, 13%). Of 520 suspected cases with response data, 184 were excluded. These included 129 young people aged ≥ 16 years at presentation or who were prevalent rather than new incident cases; 31 with ambiguous genitalia; 23 whose symptoms did not meet case validation criteria; and one where sexuality was conflated with gender identity (no GD). There were 53 erroneous notifications. 279 cases met case definition for inclusion (246 notified through CAPSS, 32 through the BPSU, and one notified by the GP), resulting in 230 confirmed cases after exclusion of duplicates.

Figure 2. Flow chart of case identification



*Duplicate notifications represent 41 cases: 34 duplicate, 6 triplicate and 1 quadruplicate notification(s).

Incidence

Of 230 confirmed cases across all countries, 57 (24.8%) were aged <12 years and 173 (75.2%) aged ≥12 years. **Table 1** shows the cases identified and estimated incidence of GD among 4- to 15-year olds presenting to secondary care or specialist gender services by country across the British Isles. Incidence across all UK countries was 1.58 to 1.80 per 100,000, lower in the Republic of Ireland (0.41 per 100,000), and higher in the Isle of Man (12.23 per 100,000).

Table 1. Cases notified and incidence of gender dysphoria by country

	Cases notified over 19-month study period			Cases notified per 12-month period	Country population aged 4-15 years, 2012 (thousands)	Incidence per 100,000 population aged 4-15 years (95% CI)
	< 12 years at diagnosis	≥12 years at diagnosis	Total			
UK^a	55	168	223	140.84	8781.2	1.60 (1.36 – 1.86)
England ^b	46	141	187	118.11	7409.7	1.59 (1.33 – 1.90)
Scotland ^c	3	14	17	10.74	678.1	1.58 (0.88 – 2.83)
Wales ^d	6	5	11	6.95	413.0	1.68 (0.81 – 3.44)
N. Ireland ^e	0	8	8	5.05	280.5	1.80 (0.77 – 4.13)
Republic of Ireland^f	2	3	5	3.16	774.5	0.41 (0.11 – 1.26)
Isle of Man^g	0	2	2	1.26	10.3	12.23 (2.35 – 54.09)
TOTAL	57	173	230			

Population sources:

a. UK: Office of National Statistics (ONS) UK Mid-Year Population Estimates 2012.

b. England: ONS England Mid-Year Population Estimates 2012.

c. Scotland: National Records of Scotland Mid-Year Population Estimates 2012.

d. Wales: ONS Wales Mid-Year Population Estimates 2012.

e. Northern Ireland: Northern Ireland Statistics and Research Agency Mid-Year Population Estimates 2012.

f. Ireland: Irish Central Statistics Office Population Estimates 2013.

g. Isle of Man: Population estimates 2013 supplied by: Economic Affairs department, Cabinet Office.

Socio-demographics and age of GD presentation

Demographic, family and school characteristics of confirmed cases are shown in **Table 2**. Assigned females comprised 57.8% of cases overall. Whilst assigned males predominated amongst those presenting with GD in <12 years (36/57, 63.2%), in adolescence, assigned females were the majority (112/173, 64.7%). The great majority were white (94%), with low representation from ethnic minority populations observed. Only 82.5% attended mainstream schools, with a history of significant bullying in 47.4%, and 4.4% (n=10) were home schooled. One in twenty (n=12) were in local authority care, with a history of a social care referral for suspected abuse in 13.3%. The majority of cases presented in adolescence (75.2%; median age 14.7 years), although 3% (n=7) presented aged <5 years and 21.7% (n=50), aged 5-11 years (**Table 3**). Median age of onset of experiencing distress related to gender was 9.5 years (IQR 5-12), with 16% (n=37) starting <5 years (**Table 3**). **Figure 3** shows the duration of experienced GD before notification. The majority (62.6%) reported long-standing desire to be the opposite sex (for ≥ 5 years before notification in 28.3%) with significant distress relating to gender present for 2-5 years in 36.1% and for ≥ 5 years in 26.5%.

Table 2. Socio-demographic characteristics of sample assessed

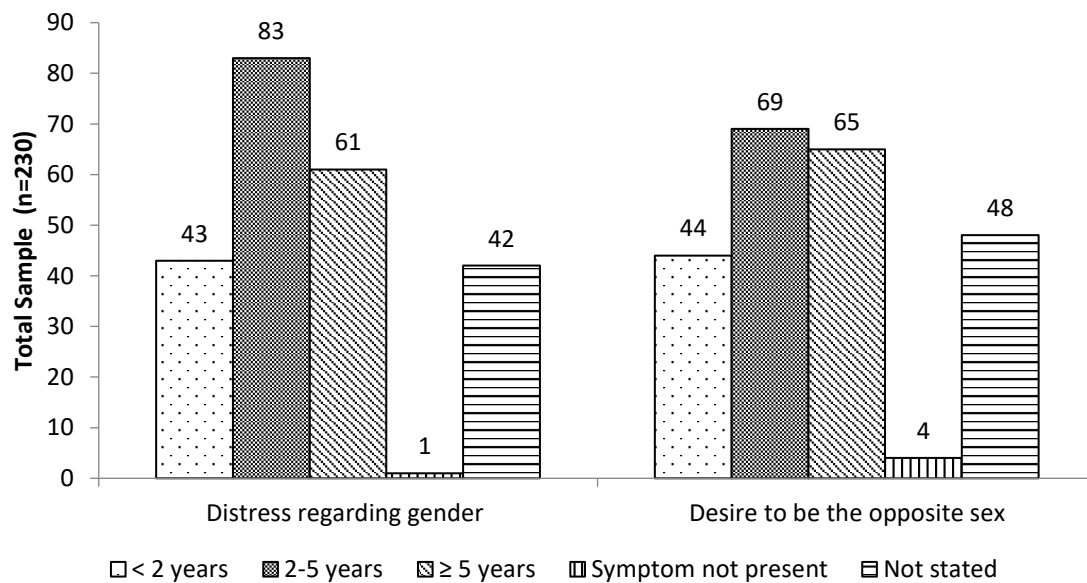
	All ages (N=230)		
	n	N ^a	%
Birth assigned sex			
Male	97	230	42.2%
< 12 years at presentation	36	57	63.2%
≥ 12 years at presentation	61	173	35.3%
Female	133	230	57.8%
< 12 years at presentation	21	57	36.8%
≥ 12 years at presentation	112	173	64.7%
Ethnicity			
White	203	216	94.0%
Mixed Ethnicity	9	216	4.2%
South Asian	4	216	1.9%
Black	0	216	0.0%
Chinese	0	216	0.0%
Other	0	216	0.0%
Family carer structure			
Single parent or reconstituted family	120	229	52.4%
Two biological parents	89	229	38.9%
Adoptive parents	4	229	1.7%
Other family member	4	229	1.7%
Looked after child (local authority care)	12	229	5.2%
History of			
Mental health disorder in parent/primary carer	69	168	41.1%
Abuse requiring Social Services referral	25	188	13.3%
Bullying requiring school action	99	209	47.4%
<95% school attendance	59	207	28.5%
Suspension or exclusion from school	15	207	7.2%
Involvement with youth offending team or other forensic service	3	215	1.4%
Schooling			
Mainstream school	188	228	82.5%
Special school	12	228	5.3%
Special unit within mainstream school	13	228	5.7%
Home schooled	10	228	4.4%
Other	5	228	2.2%

^a N= number with available data

Table 3. Age at presentation compared with age of onset of GD features among the sample assessed

Age (years)			All ages (N=230)	
	median	IQR	n	%
Age at presentation to secondary care/specialist GD service	14.68	(12.03 -15.31)	230	-
< 12 years at presentation	9.41	(7.50-10.40)	57	24.8%
≥ 12 years at presentation	14.98	(14.50 - 15.54)	173	75.2%
Age at onset of expression				
Behavioural changes	8	(4.0 – 12.0)	209	
Distress relating to gender	9.5	(5.0 – 12.0)	188	
Desire to be the opposite gender	9	(5.0 – 12.0)	179	
Age at onset of distress				
<5 years	-	-	37	16.1%
5-11 years	-	-	62	27.0%
≥12 years	-	-	89	38.7%
No distress noted	-	-	1	0.4%
Age of onset not known /stated	-	-	41	17.8%
Age at onset of desire				
<5 years	-	-	35	15.2%
5-11 years	-	-	70	30.4%
≥12 years	-	-	74	32.2%
No stated desire noted	-	-	4	1.7%
Age of onset not known /stated	-	-	47	20.4%

Figure 3. Duration of features before gender dysphoria diagnosis in the sample assessed



Co-occurring Mental Health Conditions

Mental health conditions and history of self-harm are shown by age-group in **Table 4**, alongside comparative UK data. Co-occurring mental health conditions were common, with 51.5% having ≥ 1 identified, and previous self-harm in 36.5%. The commonest conditions were depression, anxiety disorders, and ASC. The majority of neurodiversity and/or mental health difficulties were reported in adolescents, with only conduct disorder and attention deficit hyperactivity disorder more common in children < 12 years.

Service Involvement at Notification

Services children and young people were known to at notification are shown in **Table 5**. A third of children < 12 years were known to a paediatric service, relative to only 11.6% of adolescents. Three quarters of cases (77%) were known to mental health services at notification. Fifty-six percent of cases were notified by (i.e. already known to) specialist gender services. Most cases (82.6%) were known to ≥ 2 services.

Clinical Management

Table 6 shows clinical case management. In most cases (90.9%), the young person stayed open to the notifying service with an additional onward referral. No cases were discharged without an onwards referral. 28% of cases were referred onwards to a specialist gender service, bringing the total already known to or referred on to a specialist gender service to 84%. In three and six adolescent cases, respectively, onward management involved an acute paediatric or psychiatric admission.

Table 4. Self-harm and mental health conditions in young people with gender dysphoria by age

Diagnosis	All ages (N=230)			< 12 years at presentation (N=57)			≥ 12 years at presentation (N=173)			% occurring in those ≥12 years	UK population prevalence (MHCYP 2017)
	n	N ^a	%	n	N ^a	%	n	N ^a	%		
History of self-harm	81	222	36.5%	3	56	5.4%	78	166	47.0%	96.3%	
Depression	53	204	26.0%	0	53	0.0%	53	151	35.1%	100.0%	0.3% 5-10yrs 2.7% 11-16yrs
Anxiety (any disorder)	33	202	16.3%	4	53	7.5%	29	149	19.5%	87.9%	3.9% 5-10yrs 7.9% 11-16yrs
Conduct disorder	16	196	8.2%	5	49	10.2%	11	147	7.5%	68.8%	5.0% 5-10yrs 6.2% 11-16yrs
Eating disorder	7	199	3.5%	0	53	0.0%	7	146	4.8%	100.0%	0.1% 5-10yrs 0.6% 11-16yrs
Obsessive Compulsive Disorder	5	201	2.5%	0	52	0.0%	5	149	3.4%	100.0%	0.1% 5-10yrs 0.6% 11-16yrs

Attention Deficit Hyperactivity Disorder	19	203	9.4%	6	52	11.5%	13	151	8.6%	68.4%	1.7% 5-10yrs 2.0% 11-16yrs
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Autistic Spectrum Condition	30	187	16.0%	6	45	13.3%	24	142	16.9%	80.0%	1.5% 5-10yrs 1.2% 11-16yrs
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Number of mental health conditions

Any (≥1)	104	202	51.5%	15	48	31.3%	89	154	57.8%	
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None	98	202	48.5%	33	48	68.8%	65	154	42.4%	
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1	62	202	30.7%	11	48	22.9%	51	154	33.1%	
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2	28	202	13.7%	2	48	4.2%	26	154	16.9%	
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≥3	14	202	6.9%	2	48	4.2%	12	154	7.8%	
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6.1% 5-10yrs
8.2% 11-16yrs
3.4% (5-10yrs)
6.2% (11-16yrs)

^a Number with data available

Table 5. Secondary care and specialist gender services young people were in contact with at notification, by age

	<12yrs N = 57)		≥ 12yrs N = 173	
	n	%	n	%
Known to service				
Paediatrics ^a	19	33.3%	20	11.6%
<i>Paediatric endocrinologist</i>	3	5.3%	2	1.2%
Mental health services ^b	33	57.9%	144	83.2%
Specialist GD service	31	54.4%	98	56.6%
Known to single service	12	21.1%	28	16.2%
GP ^c	1	1.8%	0	0.0%
Paediatrics ^a	6	10.5%	2	1.2%
<i>Paediatric endocrinology</i>	1	1.8%	1	0.6%
Mental health service ^b	2	3.5%	21	12.1%
Specialist gender service	3	5.3%	5	2.9%
Known to 2 services	38	66.7%	131	75.7%
Known to 3 services	7	12.3%	14	8.1%

^a Paediatrics includes acute, community or specialist (e.g. paediatric endocrinology) paediatric services

^b Mental health services includes Child and Adolescent Mental Health services (CAMHS) and other mental health provision not specified as CAMHS

^c Lone case reported directly to BPSU by GP

Table 6. Management of cases meeting criteria for gender dysphoria by the notifying service, by age

	< 12YRS (N= 57)		≥ 12YRS (N = 173)	
	n		n	%
Discharged by notifying service	4		13	7.5%
<i>With onward referral^a</i>	4		13	7.5%
Ongoing follow-up by notifying service^b	53		160	92.5%
<i>Plus onward referral^a</i>	51		158	91.3%
Referrals made to/for^c:				
<i>Hormone Evaluation</i>	2		8	4.6%
<i>Paediatric Endocrinologist</i>	7		40	23.1%
<i>Child & Adolescent Mental Health Service</i>	9		9	5.2%
<i>Specialist gender service</i>	15		49	28.3%
Other management:				
<i>Paediatric inpatient admission</i>	0		3	1.7%
<i>Psychiatric inpatient admission</i>	0		6	3.5%

^aOnward referral made by notifying clinician or reported to have been made by another clinician (e.g. GP); figures may underestimate other clinician referrals

^bNo data to confirm discharge from care

^cDenotes new referrals where case not already known to the service

Discussion

We present findings from the first incidence study of GD in children and young people, using two surveillance systems. UK incidence among 4- to 15-year-olds presenting to secondary care or specialist gender services from 2011-2013 was 1.58 – 1.80 per 100,000, and similar across all UK countries. There was a lower incidence (0.4 per 100,000) in the Republic of Ireland and a higher incidence (12.23 per 100,000) in the Isle of Man. The majority were assigned females, of white ethnicity (94.0%), and presented to services in adolescence, with few notifications among 4- to 5-year-olds (3%). However, distress regarding gender preceded notification by ≥ 2 years in 62.6% and by ≥ 5 years in 26.5%, indicating onset in younger childhood in the majority, with presentation and recognition occurring late. A fifth were not in mainstream school and half had additional mental health difficulties and/or neurodiversity (51.5%), with previous self-harm in a third. Eight-four percent were known or referred on to a specialist gender service at notification.

The higher rate of notification among assigned female referrals ≥ 12 years mirrors findings among gender clinics internationally. Clinics in Toronto and Amsterdam noted a significant change in the sex ratio of referred young people between two cohort periods (2006 and 2013; Aitken et al., 2015; Kaltiala-Heino, 2020). Research by de Graaf et al. (2018) has indicated a similar shift in UK adolescent populations, compared with an equal split among younger children (<12 years), also reported elsewhere (e.g. Kaltiala-Heino, 2020). Sociological and socio-cultural explanations are offered to account for this inversion in the sex ratio of adolescents with GD (Aitken et al., 2015), but consensus has not been reached.

The current sample was predominantly of white ethnicity, mirroring previous UK research (de Graaf et al., 2019). The literature indicates that ethnic minority populations face multiple barriers in accessing healthcare services (Szczepura, 2005), perhaps due to interventions being tailored for white ethnic populations (Memon et al., 2016). The proportion of white ethnicity cases reported here is higher than among CAMHS referrals generally, as found by de Graaf et al. (2019). They suggest that feelings of stigma or shame, and cross-cultural variations in the conceptualisation of gender may act as key barriers in seeking specialist gender services.

The present study provides insight into the social circumstances of young people with GD presenting to healthcare services. Only 82.5% were in mainstream schools, with sizeable minorities in special schools/units or home schooled. Nearly half the sample (47.4%) had a history of significant bullying, as noted elsewhere (e.g. Holt et al., 2016). Whilst unclear whether this was transphobic in nature, research indicates that experience of macro-aggression and social rejection likely contribute to poor mental health in gender diverse young people (Chodzen et al., 2019; Delozier et al., 2020). Five percent of the current cohort were in care, reflecting previous UK clinic findings (Matthews et al., 2019).

Co-occurring mental health difficulties were high in this sample and may be a factor prompting referral to secondary care or specialist gender services in the British Isles. The impact of distress and nature of accompanying mental health concerns have been researched extensively among gender clinic cohorts. UK clinic data indicate co-occurring low mood in 42% and self-harming thoughts and behaviours in 39% (Holt et al., 2016; Skagerberg et al., 2013). Similar mental health difficulties have been reported in Dutch (in

32.4%, predominance of internalizing disorders; de Vries et al., 2011), Finnish (64% depression, 53% suicidal/self-harming behaviours; Kaltiala-Heino et al., 2015) and American clinic populations (Olson et al., 2015; Khatchadourian et al., 2014;). Children <12 years are more likely to present with externalising behaviours such as conduct disorder (Holt et al., 2016), replicating the current study findings. Of note, not all young people experience such difficulties with their mental health. de Vries et al. (2011) showed that 67% of young people referred to their service had no additional mental health difficulties. This may indicate that gender incongruence is more readily identified as a need in itself requiring referral in the Dutch cohorts studied.

The present findings also support previous research indicating a higher prevalence of ASC in young people with GD presenting to healthcare services (16% in this cohort) than in the general population. Holt et al. (2016) reported rates of 13.3% among UK clinic referrals compared with 9.4% and 26% among Dutch (de Vries et al., 2010) and Finnish clinic populations, respectively (Kaltiala-Heino et al. 2015). Differences may relate to the type of assessment employed: insinuated diagnosis via screening questionnaire as compared with formal diagnosis using tools such as the Diagnostic Interview for Social and Communication Disorders (DISCO).

The current study suggests that features and distress are frequently present for extended periods before GD is diagnosed. The high levels of social adversity and mental health difficulties identified in this study may indicate a need to lower thresholds and facilitate earlier referrals for specialist support for young people with GD. This might also explain why fewer under 12s were notified during the study; lower rates of mental health difficulties seen in children <12 years may influence likelihood of recognition of GD, and referral to specialist gender services, in this age-group. Further research is required to explore the apparent delay between reported onset and subsequent referral.

Strengths and limitations

The present research is the first of its kind to provide incidence figures for GD in young people aged 4 to 15 years using recognised systems. The work is particularly unique owing to its use of two surveillance systems to maximize potential for identifying relevant cases. Incident cases referred to the two UK specialist child and adolescent gender identity services, in England and Scotland, were captured through CAPSS via the psychiatrists within these services. Nonetheless, despite high uptake from paediatricians and psychiatrists across the British Isles, it is likely that incidence rates reported here are a minimum estimate. This is especially so when investigating presentations with complex case definitions and diffuse recognition of onset. Young people with significant distress or associated difficulties might be more likely to have been referred to paediatric or mental health services and thus to have been notified to the study, whereas some with lesser degrees of distress might not have presented to or been referred to health services and would not be represented in the present research. This results in particular difficulties in determining the number of pre-adolescent children who experience GD, and could delay formal assessment for some years after onset of gender distress (as shown in the presented cohort). Similarly, owing to an age cut-off of 15.9 years for inclusion in this study, the presented data under-represent the total number of adolescents experiencing GD and impact on services. Lack of family acceptance, cultural barriers, or stigma may affect access

to specialist gender services for some young people. Less awareness of, or services for, GD in some areas may also have affected estimates, for example in Republic of Ireland. Present incidence rates do not include those who have pursued private treatment and care pathways, only those referred to national child health services.

More recent evidence suggests that incidence may have risen since the present study ended in 2013. Specialist gender services in the UK and other countries (e.g. de Graaf et al., 2017; Kaltiala-Heino et al., 2020) have reported rising referral rates, although there are no data on whether this represents changing referral patterns from secondary to specialist gender services, higher rates of presentation, increased awareness of gender services, or a true increase in incidence.

The present research does not describe the trajectory of cases after initial notification. More research assessing subsequent management and outcomes of gender diverse young people is warranted to optimize care pathways and service provision for this group.

Conclusions

This is the first baseline incidence study of GD in children and young people and used surveillance methodology to identify cases presenting to secondary care and specialist gender services across the British Isles. Although rare, GD was associated with significant levels of mental health difficulties and higher rates of ASC and social adversity. The findings and long time to diagnosis raise important questions about whether co-occurring difficulties could be mitigated or reduced if this group had earlier access to specialist support.

References

Aitken, M., Steensma, T. D., Blanchard, R., VanderLaan, D. P., Wood, H., Fuentes, A., ... & Zucker, K. J. (2015). Evidence for an altered sex ratio in clinic-referred adolescents with gender dysphoria. *The journal of sexual medicine*, 12(3), 756-763.

Almeida, J., Johnson, R. M., Corliss, H. L., Molnar, B. E., & Azrael, D. (2009). Emotional distress among LGBT youth: The influence of perceived discrimination based on sexual orientation. *Journal of youth and adolescence*, 38(7), 1001-1014.

BPSU Annual Report (2020-2021). <https://www.rcpch.ac.uk/sites/default/files/2021-11/BPSU-Annual-Report-2020-21.pdf>

Chodzen, G., Hidalgo, M. A., Chen, D., & Garofalo, R. (2019). Minority stress factors associated with depression and anxiety among transgender and gender-nonconforming youth. *Journal of Adolescent Health*, 64(4), 467-471.

Clark, T. C., Lucassen, M. F., Bullen, P., Denny, S. J., Fleming, T. M., Robinson, E. M., & Rossen, F. V. (2014). The health and well-being of transgender high school students: results from the New Zealand adolescent health survey (Youth'12). *Journal of adolescent health*, 55(1), 93-99.

- Delozier, A. M., Kamody, R. C., Rodgers, S., & Chen, D. (2020). Health disparities in transgender and gender expansive adolescents: A topical review from a minority stress framework. *Journal of Pediatric Psychology, 45*(8), 842-847.
- de Graaf, N. M., Giovanardi, G., Zitz, C., & Carmichael, P. (2018). Sex ratio in children and adolescents referred to the Gender Identity Development Service in the UK (2009–2016). *Archives of Sexual Behavior, 47*(5), 1301-1304.
- de Graaf, N. M., Manjra, I. I., Hames, A., & Zitz, C. (2019). Thinking about ethnicity and gender diversity in children and young people. *Clinical child psychology and psychiatry, 24*(2), 291-303.
- de Graaf, N. M., Steensma, T. D., Carmichael, P., VanderLaan, D. P., Aitken, M., Cohen-Kettenis, P. T., ... & Zucker, K. J. (2020). Suicidality in clinic-referred transgender adolescents. *European Child & Adolescent Psychiatry, 1-17*.
- de Graaf, N. M., Cohen-Kettenis, P. T., Carmichael, P., de Vries, A. L., Dhondt, K., Laridaen, J., ... & Steensma, T. D. (2017). Psychological functioning in adolescents referred to specialist gender identity clinics across Europe: a clinical comparison study between four clinics. *European child & adolescent psychiatry, 27*(7), 909-919.
- de Vries, A. L., Doreleijers, T. A., Steensma, T. D., & Cohen-Kettenis, P. T. (2011). Psychiatric comorbidity in gender dysphoric adolescents. *Journal of Child Psychology and Psychiatry, 52*(11), 1195-1202.
- de Vries, A., Noens, I., Cohen-Kettenis, P., van Berckelaer-Onnes, I., Doreleijers, T. (2010). Autism spectrum disorders in gender dysphoric children and adolescents, *J Autism Dev Disord, 40*, 930–6.
- de Vries, A. L., Steensma, T. D., Cohen-Kettenis, P. T., VanderLaan, D. P., & Zucker, K. J. (2016). Poor peer relations predict parent-and self-reported behavioral and emotional problems of adolescents with gender dysphoria: a cross-national, cross-clinic comparative analysis. *European child & adolescent psychiatry, 25*(6), 579-588.
- Gray, S. A., Carter, A. S., & Levitt, H. (2012). A critical review of assumptions about gender variant children in psychological research. *Journal of Gay & Lesbian Mental Health, 16*(1), 4-30.
- Holt, V., Skagerberg, E., & Dunsford, M. (2016). Young people with features of gender dysphoria: Demographics and associated difficulties. *Clinical child psychology and psychiatry, 21*(1), 108-118.
- International Statistical Classification of Diseases and Related Health Problems (11th ed.; ICD-11; World Health Organization, 2019).

Kaltiala, R., Bergman, H., Carmichael, P., de Graaf, N. M., Egebjerg Rischel, K., Frisen, L., ... & Waehre, A. (2020). Time trends in referrals to child and adolescent gender identity services: A study in four Nordic countries and in the UK. *Nordic journal of psychiatry*, 74(1), 40-44.

Kaltiala-Heino, R., Bergman, H., Työläjäarvi, M., & Frisén, L. (2018). Gender dysphoria in adolescence: current perspectives. *Adolescent health, medicine and therapeutics*, 9, 31.

Kaltiala-Heino, R., Sumia, M., Työläjäarvi, M., & Lindberg, N. (2015). Two years of gender identity service for minors: overrepresentation of natal girls with severe problems in adolescent development. *Child and Adolescent Psychiatry and Mental Health*, 9(1), 1-9.

Khatchadourian, K., Amed, S., & Metzger, D. L. (2014). Clinical management of youth with gender dysphoria in Vancouver. *The Journal of pediatrics*, 164(4), 906-911.

Maughan, B., Rowe, R., Messer, J., Goodman, R., & Meltzer, H. (2004). Conduct disorder and oppositional defiant disorder in a national sample: developmental epidemiology. *Journal of child psychology and psychiatry*, 45(3), 609-621.

Meltzer H, Gatward R, Goodman R, Ford T. *Mental health of children and adolescents in Great Britain*, The Stationary Office: London, 2000.

Meltzer, H., Gatward, R., Goodman, R., & Ford, T. (2003). Mental health of children and adolescents in Great Britain. *International review of Psychiatry*, 15(1-2), 185-187.

Memon, A., Taylor, K., Mohebbati, L.M., Sundin, J., Cooper, M., Scanlon, T., & de Visser, R. (2016). "Perceived barriers to accessing mental health services among black and minority ethnic (BME) communities: a qualitative study in Southeast England." *BMJ Open*. 6(11). doi: 10.1136/bmjopen-2016-012337.

Mental Health of Children and Young People in England, 2017 (2018)
<https://digital.nhs.uk/data-and-information/publications/statistical/mental-health-of-children-and-young-people-in-england/2017/2017>

Olson, J., Schragar, S. M., Belzer, M., Simons, L. K., & Clark, L. F. (2015). Baseline physiologic and psychosocial characteristics of transgender youth seeking care for gender dysphoria. *Journal of Adolescent Health*, 57(4), 374-380.

Olyslager, F., & Conway, L. (2007, September). On the calculation of the prevalence of transsexualism. In *World Professional Association for Transgender Health 20th International Symposium, Chicago, Illinois. Retrieved April* (Vol. 22, p. 2010).

Ristori, J., & Steensma, T. D. (2016). Gender dysphoria in childhood. *International Review of Psychiatry*, 28(1), 13-20.

Skagerberg, E., Davidson, S., & Carmichael, P. (2013). Internalizing and externalizing behaviors in a group of young people with gender dysphoria. *International Journal of Transgenderism*, 14(3), 105-112.

Skordis, N., Kyriakou, A., Dror, S., Mushailov, A., & Nicolaidis, N. C. (2020). Gender dysphoria in children and adolescents: an overview. *Hormones*, 1-10.

Steensma, T. D., Kreukels, B. P., de Vries, A. L., & Cohen-Kettenis, P. T. (2013). Gender identity development in adolescence. *Hormones and behavior*, 64(2), 288-297.

Szczepura, A. (2005). Access to health care for ethnic minority populations. *Postgraduate medical journal*, 81(953), 141-147.

Thrower, E., Bretherton, I., Pang, K. C., Zajac, J. D., & Cheung, A. S. (2020). Prevalence of autism spectrum disorder and attention-deficit hyperactivity disorder amongst individuals with gender dysphoria: a systematic review. *Journal of autism and developmental disorders*, 50(3), 695-706.

van der Miesen, A. I., de Vries, A. L., Steensma, T. D., & Hartman, C. A. (2018). Autistic symptoms in children and adolescents with gender dysphoria. *Journal of Autism and Developmental Disorders*, 48(5), 1537-1548.