

BMJ Open Lifespan healthcare transitions among individuals with intersex traits in Canada: a mixed-methods and qualitative study

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ABSTRACT

Objectives To: (1) complete an integrative literature review of transition studies that focus on individuals with intersex traits; (2) conduct an environmental scan of the current resources (practice guidelines, policies and procedures) used by healthcare providers working with Canadians with intersex traits; (3) investigate the experiences of Canadians with intersex traits in their healthcare transitions across the lifespan and (4) assess the understanding of healthcare providers about these transitions.

Design A qualitative prospective community participation study was conducted. It used mixed methods including an environmental scan and semistructured engagement sessions.

Setting The environmental scan examined resources available throughout Canada. The engagement sessions took place in British Columbia and Ontario, Canada.

Participants Sixteen participants were recruited. These included 13 individuals with intersex traits (a heterogeneous group of congenital conditions affecting the development of sex characteristics) and three caregivers.

Methods Mixed methods included an integrated literature review, environmental scan and qualitative approaches developed in collaboration with community partners.

Results The literature review identified gaps in transition care for individuals with intersex traits. The environmental scan uncovered no specific resources used by healthcare providers working with patients with intersex traits, though several general guidelines were used. Engaging providers in the study was problematic. Thematic analysis generated three main themes that emerged from the engagement sessions: (1) transition is a lifespan activity; (2) building personal agency is valuable and (3) well-being promotion is an application of health literacy.

Conclusions Transition resources for individuals living in Canada with intersex are scarce. Transitions happen across the lifespan with ownership of thought and actions seen as acts of personal agency. Health literacy skills and knowledge change with increased age, yet the primary source of knowledge often remained important in the individual's autobiographical self.

Strengths and limitations of this study

- This paper focuses on the healthcare transition experiences of individuals with intersex traits in Canada specifically, which has not yet been investigated.
- This study utilises a mixed-methods approach. Specifically, community engagement recognises members' rich knowledge base, experience and values to improve its inclusive language use and inform future healthcare practices.
- Limited participation levels and an unwillingness from healthcare providers to be recorded during the engagement sessions may reflect topic sensitivities.

INTRODUCTION

As a heterogeneous group of congenital conditions, intersex variations/differences in sex development (DSD), or intersex traits, are typically noted at birth, early childhood or adolescence. This article uses the terms intersex or intersex traits, as preferred by the community informing our study as well as broader advocacy and support groups.¹ Historic and current management of intersex is complex in healthcare management. Despite attempts from early 2000 onwards to develop interdisciplinary approaches, outside of Canada,^{2–4} quality care indicators and long-term wellness outcomes remain difficult to discern. At an individual level, intersex continues to be problematised by healthcare actions and social misconceptions.⁵ Like others globally, Canadians with intersex traits experience numerous health inequalities, including irreversible (genital) surgeries largely in childhood.^{5,6} While intersex refers to a large group of conditions that affect hormones, reproductive development and/or genital appearance, a 'true' prevalence is unclear⁷ and the idea of essential intervention is contested.⁸ Diminished rights of individuals with intersex can contribute to

uncertainty and bodily frustration as a consequence of early surgical intervention,^{9 10} as well as poor specific (traits focused) and general daily-living (physical, mental and reproductive) health outcomes.^{8 9 11}

Care of individuals with intersex traits includes addressing issues of inequity and stigmatisation.^{12–14} Being born with intersex can impact physical,^{15 16} psychosocial^{16 17} and familial^{18 19} experiences at organisational,^{20 21} policy,^{22–24} cultural and community levels.²⁵ Psychosocial studies have illuminated a range of concerns for individuals with intersex linked to quality of life, trauma, mistrust, mental health concerns and isolation.^{26–28} Despite shifts in legislation globally, emerging emancipatory studies,^{20 29} human rights champions and leaders,^{30 31} and heightened public awareness of diversity,^{32 33} healthcare across the lifespan for individuals with intersex can be compromised. Furthermore, optimal lifespan management approaches remain unclear with limited outcome reporting and tension between activists and professional (healthcare) groups. Rather, much of the work to date focuses on transition at the point of exit from paediatric services,³⁴ focusing on self-management and mastery. For individuals with intersex traits the shift towards adulthood is a time of personal awareness, autonomy and responsibility in health management.¹² Youth need to assimilate the information learnt from parents and providers and navigate applying wider social concepts to themselves. Occurring at an age of immense biopsychosocial and environmental change, transition carries risks which include: deterioration in health as a consequence of withdrawal from provider contact,³⁵ self-care neglect linked to developmental maturation, increased burden of care linked to both human and financial factors, as well as lost opportunities to access education, healthcare records and peer support.

The Canadian Institutes of Health Research (CIHR) note healthcare transitions happen across the lifespan but are usually focused on times when care is transferred between providers, institutions and/or sectors.³⁶ Transition is commonly reported within the pediatric-to-adult-context and viewed through a lens of youth emancipation and self-management,³⁷ which neglects the young adult-adult and older adult life course. As individuals with intersex transition through life's trajectories, this necessitates a change in care provider from youth to adult³⁴ and health status changes because of ageing, wider social factors¹² or access to specialist healthcare services. However, little is known about the transition experiences of adults living in Canada.

Unlike other countries that have developed a research community and/or patient support (ie, DSD-life,³⁸ Germany; I-CAH,³⁹ UK; DSDnet,⁴⁰ EU; DSD-TRN,⁴¹ USA) Canada lacks formalised, charitable national intersex networks, advocacy and peer support groups offering universal healthcare guidance and provincial signposting to services (though there are examples of condition-specific parent support groups that have in the past been linked to intersex, ie, Turner Syndrome Society of

Canada,⁴² Klinefelter Syndrome Association of Canada⁴³ and Intersex London⁴⁴). Furthermore, Canada lacks provincial directives, and it is unknown how existing globally recognised quality practice standards, pathways or intersex-affirming hospital policies supporting transition across the lifespan have been applied. Moreover, there is an absence of data from adults in Canada with intersex. Their experiences of healthcare lifespan transitions are important to gather due to Canada's unique multicultural and multiethnic population as well as widespread geography. Such limitations may challenge the applicability of international resources that are often developed from engagement with primarily white and urban populations.

This article is a first step in addressing the gaps in knowledge and healthcare for individuals with intersex living in Canada. This paper first reviews literature addressing intersex and healthcare transition. The paper reports results from a broad environmental scan across Canada including a review of intersex resources and approaches that guide practice. Finally, this paper reports qualitative data investigating the experiences of healthcare transitions in Canadian individuals with intersex traits. Long term we hope to explore psychosocial supports for individuals with intersex, develop health literacy resources and explore how individual–healthcare–provider relationships can be strengthened.

METHODS

A CIHR-funded mixed-methods study, as a form of triangulation,⁴⁵ took an emancipatory approach, aiming to empower people with intersex to assess and imagine the ideals for their own healthcare transition experiences. Investigator triangulation drew on successful community-collaborations^{20 46} to better understand health transition experiences for Canadians with intersex. The study included (1) a literature review of international intersex transition studies, (2) an environmental scan from paediatric hospital sites across Canada and (3) focus groups or interviews adopting a community-participatory approach that aimed to understand firsthand experiences of transition care in the Canadian healthcare system for individuals with intersex; for detail, see study design.^{47 48 49}

Literature review

To better understand the mechanisms that facilitate transition and ongoing engagement with healthcare across the lifespan for individuals with intersex, an integrative literature review^{50 51} was conducted first in 2019 (to guide interview question development) and repeated later (to aid interpretation of findings), incorporating both qualitative and quantitative studies. The search was completed between October 2020 and February 2021 across five databases and one Journal catalogue: Medline, CINAHL, PsycINFO, Web of Science, Journal of Pediatric and Adolescent Gynecology, Google Scholar and hand searching of reference lists of included papers. A variety of key terms and their appropriate truncation

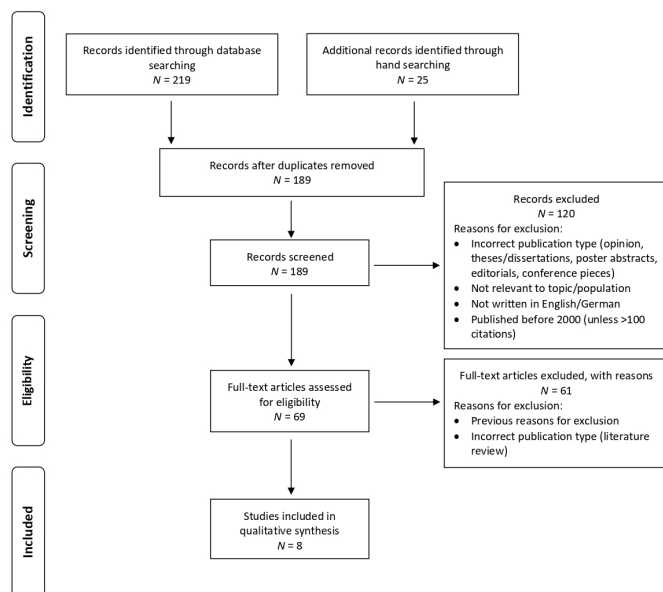


Figure 1 Literature search strategy.

depending on the database were used for the searches, linked through BOOLEAN operators (see online supplemental material table 1). Searches were limited to studies written in English/German (as we had access to a translator), published between 2000 and 2021 unless receiving greater than 100 citations. Inclusion criteria comprised of clear study method and intersex trait reporting while exclusion included opinion, dissertation, poster abstracts or conference materials. The literature review was conducted in three phases, by three authors (CS, MU and EA) using DistillerSR⁵²: (1) title and abstract screening to achieve consensus per inclusion and exclusion criteria (2) full-text screening that informed agreement of critical appraisal questions to support data extraction and (3) data extraction (figure 1⁵³).

Environmental scan

The environmental scan assessed the resources, policies, procedures or guidelines currently used by Canadian healthcare providers (HCPs) to guide care with individuals with intersex. Over a 2-month period in 2020, 13 Canadian Children's Hospital websites were searched for resources and policies available online, regarding healthcare transitions throughout the life course for persons with intersex. Key words used within the hospital website search bars included: 'transition', 'intersex', 'difference/disorder sex development', 'sex variation', 'Turner syndrome', 'congenital adrenal hyperplasia' (CAH), 'androgen insensitivity syndrome', 'Klinefelter syndrome' and 'hypospadias', as well as spelling and abbreviated variations.

Informed by the literature review and environmental scan of hospital websites, a brief anonymous 11 question HCP survey was developed (see online supplemental material table 2). A consent paragraph was provided in the introduction email. The survey was distributed to over 500 email addresses via peer networks and providers

identified on hospital websites and through professional societies.

Due to poor survey response rates, freedom of information requests (FOIs) costing a total of \$C130 were submitted to each of the 13 Canadian Children's hospitals in February and March 2020 requesting evidence of resources and policies including but not limited to: practice guidelines and policies surrounding the care of individuals with intersex, procedures regarding infants, children or youth with intersex (see online supplemental material table 3). Responses were received 1–7 months later (see <https://doi.org/10.5281/zenodo.6092189>)

Engagement sessions

Canadians aged 16+ years with intersex, caregivers or HCPs were targeted through a combined recruitment campaign including: word-of-mouth, active advertisement including local lesbian, gay, bisexual, transgender, queer and intersex (LGBTQI+) magazines and websites, unpaid Facebook and Instagram platforms, and via intersex organisations (Intersex Canada,⁵⁴ Organisation Intersex International⁵⁵) in British Columbia (BC) and Ontario (ON). HCPs were also recruited through networks, professional groups and conference postings across BC and ON. This strategy was chosen given a lack of organised networks with which to sample from for this population. As a result of the sampling strategy, no reliable estimate of either group of individuals could be calculated.

Meetings and interviews were semistructured, all participants provided written informed consent, and an honorarium was provided. Informed by the first literature review, questions discussed individuals' understanding and experiences of transitions in care, and their recommendations for future transition practices (see online supplemental material table 4). Interviews were audio recorded with participant permission. The study team were not involved in any of the participants' care. Interviews were transcribed by an independent third party. Researchers coded transcripts and conducted thematic analysis in ATLAS.ti,⁵⁶ following Braun and Clarke's method for identifying, analysing, organising, describing and reporting themes.⁵⁷ An audit trail, team discussion and reflexivity were central to analysis.⁵⁸ Themes were discussed until consensus was reached.

Participant demographics

Sixteen individuals were interviewed. In-person stakeholder engagement meetings with five participants, two individuals, and three caregivers, were conducted at a city in BC in October 2019 in groups of either two or three (FG1, FG2). From March 2020 to October 2020, interviews were individually conducted online due to the COVID-19 pandemic. Of these, two BC participants were individually interviewed via Zoom (P1, P2) and two participants from ON were interviewed individually, one via telephone (P3) and one via Zoom (P4 asked not to be recorded). Ethical approval (E2017.0922.068.00) was

then obtained to use data collected between 2017 and 2019 via interviews or workshops to inform knowledge translation outputs (KT) from seven Canadian women with CAH (P5–P11), since transition was a topic in relation to healthcare experiences. No demographic data nor diagnosis data was purposefully recorded to maintain anonymity; all disclosed to interviewers their individual diagnosis and age, with permission sought to include these data. As there is a wide range of intersex traits, all participants disclosed a diagnosis that is described under the current nomenclature.

Despite reaching out to 41 Canadian professional organisations supporting individuals with intersex, only two informal discussions occurred with HCP one from BC and one from ON; however, there was no consent to record nor take notes, thus these data are excluded.

Patient and public involvement

One individual from a prior study focused on CAH and one patient partner provided advice and informed study design. Their input was taken into consideration for final decision making. Neither wished to be identified on the study proposal, manuscript or at presentation of any findings. Furthermore, we have also developed websites^{48 49} to help with dissemination of research and knowledge to the public.

RESULTS

Literature review

Eight relevant articles were uncovered (online supplemental material table 5). Articles included audits,^{33 59} cohort studies^{60–64} and one case series study.⁶⁵ Studies assessed a range of factors in relation to transition, including the status of patient transition needs,^{33 61} engagement with, and access to adult services,⁵⁹ satisfaction with a specific transition programme,⁶⁰ perceived preferences and barriers to various services (eg, transition programmes),⁶⁰ individual and parent concerns across the lifespan,^{65 66} adult outcomes⁶² and individuals' experiences of healthcare transition.^{64 65} No Canadian studies were identified.

Environmental scan

From the search across the 13 Canadian children's hospital websites, several resources related to either intersex or healthcare transitions were uncovered. However, no resources related to both intersex and healthcare transition.

The survey garnered three responses (see online supplemental material table 2) despite over 500 emails being sent. One respondent used transition materials for pediatric-to-adult care. One used discharge summary resources with individuals with intersex.

From the FOI requests sent to each Children's hospital, two hospitals reported using practice documents (see online supplemental material table 3). Eleven reported

no policies, guidelines, or procedures for working with individuals with intersex.

The results of both the literature review and the environmental scan outline the lack of evidence-based and formalised resources, policies, procedures or guidelines used by HCPs when working with individuals with intersex traits in Canada, as well as the lack of resources that are available to these individuals when searching hospital websites for information about their conditions.

Engagement sessions

The following sections examine the three themes dominant across the engagement sessions data, providing excerpts shared verbatim to illustrate impact and high-light metaphors of significance.

Theme 1: transition is a lifespan activity

First, transition was constructed by participants as a lifespan activity encompassing the whole life course, despite transitions in care often being focused on the pediatric-to-adult care experience. Individuals in our study reported facing challenges across their life course, which became part of their changing journey as they aged. Sourcing different providers because of ageing was a common challenge, with the role of a primary care provider becoming central in helping individuals navigate healthcare complexity outside of intersex '...the intersex or the AIS [androgen insensitivity syndrome] or whatever takes a real background to gerontology, I'm 77,...dealing with sort of end-of-life issues' (P1).

Changes in care meant that individuals needed to proactively seek out new healthcare partnerships, which for some included letting go of trusted relationships with specialists. When individuals moved geographically, as noted by FG1, there was a stepwise approach or sequence in events that brought about opportunity to make new healthcare relationships:

...like transitioning from like a family doctor who I had since I was a baby to my endocrinologist [in adolescence] when I was in whatever grade to moving here [new city] to then having [to find a new] family doctor so.....transitions within those practitioners and the various experiences (FG1).

Geography transitions were also connected with immigration to Canada, which resulted in a new medical team and new diagnosis (P4). A prior misdiagnosis or curiosity about an individual's diagnosis resulted in transitions in self that were unexpected for P4, while P3 noted neglecting exploring a diagnosis, 'since I was too busy.' P2 argued early relationships with specialist providers supported a transition to similar specialists:

[The Specialist] didn't speak to me like I was a child at any point [but] like we were very much equals... and I feel like very much the same now with the adult endocrinologist, but I don't know if that would be like a universal thing (P2).

For P2, establishing a satisfactory primary care relationship took a little more effort; '(the) nurse practitioner, that I kind of see as my family doctor, set me up with an online service that doesn't actually have my records.' For others, there was an expectation that transition also meant a responsibility to have an acceptable level of knowledge about their healthcare needs, as P5 noted, 'I should know these things, being 40.' Further discussion focused on transitions not only linked to age but also to life events such as motherhood. A request to visit genetic counselling and a requirement to attend prenatal care and connect with new specialist providers who knew less about steroid management resulted in uncertainty:

...gestational diabetes's twice, nobody told me that [my condition] affects your blood sugars. Not until I got gestational diabetes and it's basically, oh, by the way this is because of your condition (P5).

Transition then became focused on a reluctance to let go of daily glucose testing, worrying about well-being, and finding courage to seek out community care, as stated by P5, 'you kind of have to seek it out yourself. Nobody refers you out.' For others, the notion of transition was linked to bodily functionality with age, how a specific condition can influence hormones and limit future fertility, 'down the road,' (P7) or, 'declining hormones,' resulting in a less visible genital difference (P3). P7 discussed celebrating moments when the body gave signals that there was hope for a future transition to motherhood; 'at 27 I go months with nothing, or I'll go months with continuous spotting and then I'll ovulate like once in a while and I'll go whoo hoo at least I'm still ovulating' (P7).

For some, transition related to uncertainty. Despite taking medication as prescribed, attending for blood work, and following medical advice there was a space in which the body 'transitioned' between being in harmony with medical care or out of sync, as described by P6:

Almost being 30, I'm really not old yet, but I'm not in my teens anymore...The Dr. is concerned about my bloodwork. In the past year and a half, mainly my steroid dose, we've been really trying to get that under control. For some reason, we haven't been able to get that quite right [even though] we've been playing with the dosage (P6).

Transition as a life course event was inherently linked to being relational, with others and self. This changed for individuals over time as they aged, were able to find independence, and confidence in understanding their bodies. There remained a desire for support especially in the transition spaces and phases within adult care with FG2 agreeing that, '(you're) on your own' which is significant since FG1 concisely stated, 'it's a life of transitions.'

Theme 2: the building of personal agency

Second, participants emphasised the value in building personal agency over the life course. Personal agency focused on the ways in which individuals had the capacity

to act, both towards self-care and knowledge with confidence, throughout transition phases. For some participants, awareness of being intersex started in childhood which continued until present day as adults or older adults. For others, there was low-agency, that is, little power or control over their feelings or actions since they had knowledge gaps about their body from childhood, 'I have no idea what the purpose of this [genital examination] was,' only later connecting this with being, 'big enough 'down there' for intercourse,' (P5). When the diagnosis of intersex came during adolescence, there was a sense of drowning in information, needing to process it quickly but being unable to do so effectively and with self-compassion. For example, participants said, 'You're still downloading a lot of information about yourself and realising that you're different and what is that difference and how does that resonate with you and everything?' (FG1); and '[t]here was a lot of things I didn't understand about my condition and my understanding was so limited that I didn't even know what questions to ask' (P8). A diagnosis in young adulthood was also hard, with P1 saying over 50 years ago, 'I was sort of hoping it wasn't true' and:

I'm sort of sugar coating because I sound as though I had it all together and I must admit that I thought I had it all together when I was 25. I thought well, this is it, this is what's gone down and then it didn't hit me until I was 29 and ... I was just unhappy (P1).

Constructing a healthy level of agency, or even countering perceived over-agency from others, that is, those with good intentions or overly caring approaches, was difficult for adolescents growing up aware they were intersex. As P5 reflected, 'not sharing everything with mom and dad...or the doctor' was a way to learn about holding 'privacy' and managing anger. Emotions were reflective of individuals' states of mind at various stages in their lives described as 'emotional health,' (FG1) which influenced relationships with self and others. While there was no specific time, transition experience, or event that triggered emotional awareness or growth, it was talked about by most participants as being in their mid-to-late-twenties and early-thirties. Recognising, reflecting, and attending to emotions was also linked to personal agency, time, resources and supports:

...people who have intersex problems, they need therapy when they're ready...it has to be when you're actually in a position to be able to deal with it and then it has to be somehow available and somehow recognized (P1).

The focus groups allowed for a general discussion of the harm misinformation had. For example, some individuals believed they 'couldn't have sex at all,' (FG2) which generated dialogue around the idea of information literacy and sex education in schools. One participant reflected on how their loss of confidence was linked to school education being 'very heteronormative and not very informative' (FG2).

Developing agency was also possible because of peer-to-peer networking and support groups. Those with CAH did not connect with others with CAH while the remaining participants accessed support groups, conferences, or online forums. Such spaces allowed individuals to use language to describe themselves, 'saying [at the] conference, they have AIS, DSD but [not] using it outside of that context,' (FG2). Overwhelmingly, once an individual decided to access peer support there were positive outcomes after initial hesitations; 'I wasn't sure whether I was even in the right place [support group meeting], whether... I had this feeling, am I intersex enough?' (FG2). Building personal agency took time and emotional investment which in combination had the potential to support self-enquiry; 'I feel like now as an adult a great deal of curiosity about it' (FG1), which in turn developed well-being.

Theme 3: promotion of well-being as personal health literacy

Finally, the concept of well-being was framed indirectly within day-to-day life, living well and appreciating self and others. Well-being was informed by personal health literacy, that is, being able to find, understand and use information to guide health-related decisions and actions. A critical aspect of being well was linked to sourcing health information that was 'trustworthy,' and at an 'understandable level' (FG1). This included information about daily activities such as diet, exercise, and sleep. The need for rest to nurture wellness was reported in a physical sense; 'I'll sleep for hours and hours and I still don't feel refreshed in the morning,' (P7). Wellness was also, for several participants, linked to a history of difficulties with mental health or the combination of medication and physiology as described by P8; '(I was) struggling with my blood pressure.....I saw a cardiologist [but it was] the anti-depressants,' that impacted sleep. Participants were keen to understand how to be healthy, recognising they had knowledge gaps which confounded their ability to determine which aspects of public health messaging applied to them. This was important as individuals transitioned across the life span, as they developed new conditions such as diabetes, hypertension, depression, anxiety, osteoporosis, coagulation conditions, obesity and other chronic conditions. A key aspect of being well was linked to education:

...education is the most important thing and there's going to be, unfortunately many people who are not going to be educated, you know, I want to sort of acknowledge my privilege and so to say, you know, well people should be able to educate themselves (P1).

Being able to seek, access, and critique information is also dependent on the ways in which it is presented; its complexity and inference should not be 'offensive' (P1). Having the capacity to check information was also part of staying well because, 'there's room for error too when you learn things yourself, right. Like you might not be getting the right information' (P9). When there were

uncertainties that is, how medication influences physical wellbeing, health became a 'mystery' (P9). P3 discussed recognising that the health landscape changes at a rapid pace, with new generations of doctors acknowledging both complexity and uncertainty with, 'new information being added to the systems every single day.' Yet for many, there remained frustrations when participants wanted to be educated by others, not be 'the educators' themselves.

I don't want to have to like re-teach somebody and I think that's where a lot of the frustration or like upset comes from is when you have to re-educate somebody who's supposed to be educating you. So I'm supposed to, to take care of me?...I need to find someone who has the answers...so finding my community it's definitely been helpful...to like guide you through what you need to do (P2).

Relationships with providers were an important part of well-being. Some providers had greater capability than others; 'I've also had a lot of really positive experience with healthcare practitioners,' (FG1). Those who were responsive were noted as helpful; '(...) it was really great to have these questions (...) and say [to my community] I asked this and got this back' (P2). Others lacked education and connection, 'gave terrible information' (P3) or left participants 'in the dark about my condition' (P8). Interactions with providers thus could either discuss (un) wellness in ways that supported individuals to secure a sense of well-being or result in frustration and uncertainty.

DISCUSSION

Each aspect of our study highlighted a dearth of resources, expertise and community-driven supports for individuals living in Canada with intersex, which has resonance with Garland *et al.*'s⁶ UK study. Protocols are often driven by medical models centred on intervention, which can be problematic since the evidence to inform care is increasingly legally and ethically contested both outside of^{8 23} and inside Canada.³¹

Participants imagined transition in healthcare as a lifespan activity, not just a pediatric-to-adult transfer. Our sample, while small, has similarities with USA,⁶⁶ Australian⁴⁶ and European studies³⁸ in that complex physical health challenges changed as individuals aged, and priorities shifted as they became more aware of their underlying condition. Transitions in care, at any age, are consistent with existing literature; lack of follow-up by specialist⁵⁹⁻⁶² or primary care,¹² limitations in person centred care,^{8 15 20} or access to psychosocial and psychological care.⁴⁷

Agency in our study referred to participants' knowledge, desire, and ability to act with ownership in ways that thoughtfully reflected their feelings, emotions, and subsequent actions which in turn influenced their mental health and well-being. Social aspects of normativity were present, yet often linked to the gender binary, similar to the work by Crocetti *et al.*⁶⁶ General well-being included the importance of day-to-day living and ageing, which

presently Grimstad *et al.*¹² suggests health systems are ‘woefully unprepared for’ (p.1) or trained for.¹³ Like others,^{26 27} mental health⁴⁷ was influenced by feelings of control, which was diminished when individuals reported low-agency or over-agency.

A lack of accessible Canadian information at the health-care sites reviewed, that is, Children’s hospitals, in combinations with a limit of peer networks, runs the risk of limiting opportunities that could enhance health literacy in this population. Interactions with HCPs that uplifted personal agency promoted holistic wellness, which resonates with the scope of clinical advocacy as describe by Indig *et al.*⁶⁷ Peer-to-peer connections are presently limited in Canada, yet both specialist and primary HCPs, professional associations, or charitable organisations, could look to address this.

Key recommendations for intersex care in Canada focus on addressing the gaps in healthcare, research, and legal and ethical standards. While we recognise that multidisciplinary teams (MDTs) exist across Canada, difficulties with engagement were disappointing. Perhaps the obstacles with streamlining ethics reviews across Canada in child health⁶⁸ translates to healthcare delivery models, provincial financing and priority setting. We recommend that collaborations be nurtured, and standards developed and agreed on between individuals with intersex traits and MDTs. Professional societies may have a role in developing position statements, focusing on a unification approach that could support Human Rights approaches.^{8 31} Addressing the gaps in education and training of HCPs, as well as adopting a lifespan approach, have the potential to enhance care and informed decision-making as noted elsewhere, including in other community-engaged research¹² see tips and actions for example Indig *et al.* and others^{67 69 70} Studies in psychosocial and psychological training approaches for healthcare professionals working in this field are needed, especially as the population with intersex remains largely ‘quiet’ in Canada.

Study limitations include the small sample size and a lack of HCPs’ data—issues perhaps stemming from the divide between medical and community/rights-based perspectives on intersex healthcare,²⁰ the triggering/difficult nature of revisiting problematic interventions for all stakeholders (patients, caregivers and HCPs), or the lack of training and education specifically around intersex traits. While healthcare stakeholders were approached from across Canada, individual recruitment was based only within two provinces and dependent on response to a general recruitment approach. The lack of a Canada-wide network, or even provincial networks, limits access to stakeholders. The wide variability in diagnosis and impact limits study conclusions as different intersex diagnoses generate disparities in individual perceptions and fertility outcomes.

Contributors Conceptualisation: CS, TJ and NC, patient participants; funding acquisition: CS, TJ, NC, LC, NC, J-PC; literature review: CS, EA and MU; data collection (environmental scan): CS, MU, EA and EK; organisation and recruitment

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Supplementary materials: Lifespan healthcare transitions among individuals with intersex traits in Canada: a mixed methods and qualitative study

Table 1. Literature search methods for studies assessing healthcare transitions for individuals with intersex variations.			
Date	Bibliographic Database	Search String	Limiters
October 13, 2020	Medline	MeSH Search: ((MH "Transitional Care+") OR (MH "Stakeholder Participation") OR (MH "Patient Participation") OR (MH "Health Care Quality, Access, and Evaluation") OR (MH "Health Services Accessibility") OR (MH "Outcome Assessment, Health Care")) AND ((MH "Intersex Persons") OR (MH "Disorders of Sex Development")) Results: 20 General Search: (DSD OR "difference* sex development" OR "variation sex development" OR "disorder* sex development" OR intersex) AND (transition OR "life course" OR "long term outcome" OR "health care access" OR engagement) AND (program* OR service* OR access OR health OR care) Results: 55	Date: 2000-2020 Language: English (if applicable) Human studies only
October 13, 2020	CINAHL Complete	MeSH Search: ((MH "Transitional Care+") OR (MH "Stakeholder Participation") OR (MH "Patient Participation") OR (MH "Health Care Quality, Access, and Evaluation") OR (MH "Health Services Accessibility") OR (MH "Outcome Assessment, Health Care")) AND ((MH "Intersex Persons") OR (MH "Disorders of Sex Development")) Results: 1 General Search: (DSD OR "difference# sex development" OR "variation sex development" OR "disorder# sex development" OR intersex) AND (transition OR "life course" OR "long term outcome" OR "health care access" OR engagement) AND (program# OR service# OR access OR health OR care) Results: 18	Limiters: Date: 2000-2020 Language: English (if applicable) Human studies only
October 13, 2020	PsycINFO	APA Psych Index Term Search: ((DE "Life Changes") OR (DE "Psychological Engagement") OR (DE "Health Care Access") OR (DE "Treatment Outcomes") OR (DE "Life Span")) AND ((DE "Intersex Conditions") OR (DE "Genital Disorders") OR (DE "Sex Chromosome Disorders")) AND ((DE "Health Care Services") OR (DE "Health Care Access"))	Date: 2000-2020 Language: English Human studies only

		Results: 0	
		General Search: (DSD OR "difference# sex development" OR "variation sex development" OR "disorder# sex development" OR intersex) AND (transition OR "life course" OR "long term outcome" OR "health care access" OR engagement) AND (program# OR service# OR access OR health OR care)	
		Results: 27	
October 13, 2020	Web of Science	General Search: (DSD OR "difference* sex development" OR "variation sex development" OR "disorder* sex development" OR intersex) AND (transition OR "life course" OR "long term outcome" OR "health care access" OR engagement) AND (program* OR service* OR access OR health OR care)	Date: 2000-2020 Language: English
		Results: 65	
October 13, 2020	Journal of Pediatric and Adolescent Gynecology	General Search: ("DSD" OR "di! sex development" OR "variation sex development" OR "intersex") AND (transition OR "life course" OR "long term outcome" OR "engagement" OR "health care access")	Year(s): 2000-2020 In this journal or book title: Journal of Pediatric and Adolescent Gynecology
October 15, 2020 to February 19, 2021	Google Scholar & reference lists of included articles	Hand Search: Search terms varied Results: 29	N/a

Table 2. Survey responses from healthcare providers ($N = 3$) regarding the use of various resources and services in their practice with individuals with intersex variations.

Request	Results			Open-ended answers
	Yes	No/None	N/A	
What best describes your current healthcare situation?	--	--	--	Hospital-based adult provider setting, regulator, and pediatric endocrinologist
Guidelines and/or care-pathways used in practice settings	0/3	3/3	--	--
Local or provincial protocols and/or practice standards used	0/3	3/3	--	--
Screening tools (e.g., decision-support tools, mental health, and wellbeing measures) used at clinic visits	0/3	3/3	--	--
Specific transition materials for pediatric to adult care	1/3	2/3	--	--
Discharge summary resources for patients following health care visits	1/3	2/3	--	--
Support groups or sign posting information shared at health care visits	--	1/3	2/3	--
Use of telehealth to connect with this population	--	3/3	--	--
Specialists involved in working with this population	--	--	1/3	Endocrinology, Gynecology, Genetics, Psychology, Urology
ICD-10 codes used in practice?	--	--	1/3	2590, 758.8, 752, 255.2, 255.9, 752.4, 752.6, 752.7, 752.8, 752.9
Duration organization keeps medical records	--	--	1/3	10-20 years; >20 years
Ongoing/planned research in area of VSD within organization	--	1/3	1/3	In development (1/3)

Abbreviations: VSD= Variations in Sex Development

Table 3. Results from freedom of information requests (FOI's) submitted to 13 Canadian Children's hospitals

Request	Results
<ul style="list-style-type: none"> practice guidelines and policies surrounding the care of individuals with intersex differences in sex development (DSD). procedures regarding infants, children, or youth with intersex/DSD See https://doi.org/10.5281/zenodo.6092189 for complete costing and hospital sites 	<ul style="list-style-type: none"> 11/13 hospitals returned no guidelines, policies, nor procedures. <p><i>General policies/guidelines/standards followed by HCPs at two hospitals:</i></p> <ul style="list-style-type: none"> The World Professional Association for Transgender Health (WPATH) guidelines [1] Endocrine Treatment of Gender-Dysphoric/Gender-Incongruent Persons: An Endocrine Society Clinical Practice Guideline [2] The Ontario Human Rights Code [3] Consensus statement on management of intersex disorder [4] Clinical Guidelines for the Management of Disorders of Sex Development in Childhood [5] Global Disorders of Sex Development Update since 2006: Perceptions, Approach and Care [6] Society for Endocrinology UK guidance on the initial evaluation of an infant or an adolescent with a suspected disorder of sex development (Revised 2015) [7] Caring for individuals with a difference of sex development (DSD): a Consensus Statement [8] Pediatric Decision Making and Differences of Sex Development: A Societies for Pediatric Urology and American Urological Association Joint Position Statement [9] Monthly multidisciplinary meetings variable involving Medical Genetics, Cytogenetics, Laboratory Medicine, Pediatric Endocrinology, Pediatric Gynecology, Pediatric Urology, Social Work, Sexual Medicine, Nursing, and other interested parties Monthly Royal College-accredited review of topic of interest

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Table 4. Interview questions posed to individuals with intersex variations in our engagement sessions and interviews, regarding healthcare transitions.

Interview Questions

- 1) What are ‘transition’ points in the life-cycle?
- 2) What expectations do you have at these transition points (from health care?), what does good transition care ‘look like’?
- 3) How has or will transition care support your health condition, functioning and well-being?
- 4) Which services have you engaged with to date OR you think you will involve with as you age?
- 5) Which services do you think we should provide at times of transition? – how would you want to / or do you access services?
- 6) How do you think we would know these are in place, are of good quality and use up-to-date information?
- 7) What are the differences between transition care (at any stage) for individuals living in urban and rural settings?
- 8) Are health care services that you access effective or efficient – how do you decide this?
- 9) At times of transition (meeting new providers, being referred to new services) how do you receive information about health protection, health promotion, disease prevention?
- 10) Where would you / who would you connect with if you believe that transition care is missing from your health care, or you would want to access services that meet your transition needs?
- 11) Is transition care about access to the right services and people?
- 12) How do you think we can in the future implement/ change transition care? Who needs to be involved?
- 13) In the future if you could design, plan or monitor transition care – what would it look like?

Table 5. Integrative literature review results (*N* = 8 studies).

Author, Year, Country	Study Design	Participant Characteristics				Key Methods and Findings
		N	Condition	Mean Age	Sex (F)	
Bachelot et al., 2017 <i>Country:</i> France	<ul style="list-style-type: none"> • Prospective • Cohort • Quantitative 	73	CAH (100%)	32.7y (<i>SD</i> = 5.7y; 24 – 44y)	66%	<p>Analysed effect of transition process and regular medical follow-up on health status and quality of life.</p> <ul style="list-style-type: none"> • 48/59 patients who were transferred to adult care had regular medical follow-up in adulthood. • 1/14 patients who were not transferred had a regular medical follow-up in adulthood • Patients with successful transition (regular follow-up in adult services) had better physical and psychological health, and quality of life. • Note: individuals with successful transition also had higher SES
Callens et al., 2020 <i>Country:</i> Netherlands	<ul style="list-style-type: none"> • Retrospective • Cohort • Mixed-methods 	18	TS (50%) KS (22%) CAH (11%) cAIS (11%) MRKHS (6%)	~19y (16-21y)	78%	<p>Used qualitative narrative analyses of interviews, and quantitative descriptive and questionnaire methods to determine information needs of adolescents in transition care.</p> <p><i>Interview:</i></p> <ul style="list-style-type: none"> • Adult hospital environment was more depersonalized – some wish they could have stayed with their pediatric team for longer • Psychological care was important for almost all participants, and HCPs should address psychological factors related to their feeling of being different, as well as shared decision making • Desire to receive person-centred care, while also having knowledge on the condition more broadly • Most participants would like a HCP who is, “... open, positive, and sensitive, and to provide reliable information in clear and uncomplicated language,” (p. 10) • Would like HCPs not to avoid topics such as sex/fertility <p><i>Questionnaire:</i></p> <p>Largest gap scores (ideal care minus current care) for:</p> <ul style="list-style-type: none"> • “Provides opportunities for me to meet other young people” (p. 11) • “Provides appointments that are convenient for me (and my family)” (p.11) • “Providers from child and adult care who arrange joint appointments/consulting hours for adolescents” (p. 11)

Gleeson et al., 2013 <i>Country: UK</i>	<ul style="list-style-type: none"> • Retrospective • Audit • Cohort • Quantitative 	53	CAH 21-OHD, SW (75%), SV (23%), 17-OHD (2%)	<i>Median</i> = 25.5y (18.4- 47.8y)	60%	<p>Evaluating successful transition from pediatric to adult care and effect of a Young Person Clinic (YPC) where a patient is appointed to an endocrinologist.</p> <ul style="list-style-type: none"> • 36/53 referred from pediatric to adult service, but 50% were lost to follow-up (two were never offered an appointment) • Only 19/34 attended their first new appointment and the subsequent appointment • No difference in engagement with adult services between those who attended YPC and those who did not. • Being offered more appointments during 2-3 years of pediatric service before transfer was associated with not being lost to follow-up for 3≤ years.
Godbout et al., 2012 <i>Country: France</i>	<ul style="list-style-type: none"> • Prospective • Cohort • Quantitative 	73	CAH (17%) HH/GHD (83%)	<i>M</i> = 24.7y (<i>SD</i> = 4.5y)	Total: 33%; CAH: 69%	<p>Assessing specific needs of young adults with CAH (among other endocrine conditions) to inform transition of care, reported on clinical discussions at adult clinic</p> <p><i>CAH-specific results:</i></p> <ul style="list-style-type: none"> • Discussions at initial visit to adult clinic for (percentage of) individuals included: height (92%), weight (77%), bone health (85%), patient relationships (69%), sexual activity (85%), menstruation with genital examination and breast examination (89% and 78% for female patients), reproduction/fertility (69%).
Lee et al., 2012 <i>Country: US</i>	<ul style="list-style-type: none"> • Retrospective • Case series • Qualitative 	6	<i>P5:</i> Gonadal DSD/ gonadal dysgenesis	Infancy to 32y	<i>P5: F</i>	<p>Study used six case studies to display factors related to problems in adulthood. However, healthcare transition was discussed explicitly for only one patient:</p> <ul style="list-style-type: none"> • <i>P5:</i> "She recognized an issue with transition of medical care from pediatrics since no doctor since has had a full picture of her situation or taken time to learn it. She has been unable to find a health care setting that provides the overall perspective for her diagnosis that she had as a child and teenager." (p. 146)
Liao et al., 2010 <i>Country: UK</i>	<ul style="list-style-type: none"> • Prospective • Audit • Cohort • Quantitative 	50	cAIS/pAIS (34%) CAH (32%) XY gonadal dysgenesis (18%)	17.8y (13-20y)	100%	<p>Clinicians assessed needs of patients at time of transition from pediatric to adult services using traffic light system.</p> <ul style="list-style-type: none"> • 15 (30%) categorized as green (low level of professional concern) • 21 (42%) categorized as amber (potential reason for concern) • 14 (28%) categorized as red (high/urgent level of clinical concern) • Of the clinical requirements (broadly, endocrine management, gynecological/urological requirements, and psychological support), 13/14

			Other (16%)*			individuals with red status had psychological issues (for 11/14 this was their only present issue).
Simoes et al., 2017	<ul style="list-style-type: none"> • Prospective • Cohort • Quantitative 	19	MRKHS (100%)	(10 - 19y)	100%	<p>To assess difference between healthcare preferences and current care for a range of health care services, including transitional programs.</p> <ul style="list-style-type: none"> • Though items from ‘transition programs’ had some of the lowest gap scores (i.e., difference between best practice/priority score and current care score on a scale of 7) compared to other elements of care; some of the highest gap scores within the ‘transition programs’ topic were: <ul style="list-style-type: none"> ○ ‘Assistance with search for psychological support’ (gap score = ~5/7), and ‘the hospital (center) has a dedicated space/room, in which MRKHS patients can get together’ (gap score = ~4.5/7).
Country: Germany						
Sutton et al., 2005	<ul style="list-style-type: none"> • Prospective • Cohort • Qualitative 	97	TS (100%)	(7 – 59y)	100%	<p>Interviewed individuals with TS across four age groups (childhood, adolescence, adulthood, mature adulthood), as well as their parents about their concerns related to TS, and strengths/weaknesses of healthcare.</p> <p><i>Individuals with TS: (N = 97)</i></p> <ul style="list-style-type: none"> • Infertility and stature were main concerns • Social aspects of sexual development and function, and health were secondary concerns <p><i>Parents: (N = 21)</i></p> <ul style="list-style-type: none"> • Concerns were primarily around health/healthcare (E.g., worried about making decisions around medication that could harmfully impact their child later) • Financial expense of condition was also a concern unique to parents <p><i>Recommendations for HCPs:</i></p> <ul style="list-style-type: none"> • Early diagnosis, offering hormonal interventions, increased education for all parties, strengthening physician-patient communications, increased sensitivity, compassion, and encouragement
Country: US						