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Title: Involving individuals with disorders of sex development and their parents in exploring new models of shared learning: Proceedings from a DSDnet COST action workshop

Abstract

Background: The level of connection between health care professionals and people who experience a condition that affects sex development is variable. These people and associated support groups need to be included in discussions about research and healthcare delivery.

Aim: Understanding the experiences of individuals with DSD their parents, health care providers and support groups.

Method: Workshop planning, preparation, delivery and evaluation involved members of working groups from the COST Action DSDnet. A coordinator, in collaboration with a support group representative, led workshop design and delivery.

Results: Our successful, facilitated workshop involved 33 attendees from nine EU countries. The workshop provided individuals with DSD, parents, advisory groups and professionals an opportunity for shared learning. Outputs focused on seven key areas including; diagnosis, childhood, and transition to adult care as well as fostering discussion around registries, future research topics, consent processes, and information needs across the life course. The importance of trustworthy and knowledgeable providers, time to understand such rare conditions and the place support groups have in a life-course approach were valuable learning points for all attendees.

Conclusion: Workshops can be designed and delivered in meaningful ways for all those involved in care of individuals with rare conditions.

Introduction

Conditions affecting sex development are a group of rare conditions that are often manifested in early life by an alteration in the appearance or function of the organs involved in sex development¹. The global change in discussions around the management of these conditions, termed as differences or disorders of sex development (DSD) that was initiated over a decade ago¹, has led to a greater focus on quality of life^{2,3}, interdisciplinary healthcare provision⁴, decision-making and cosmetic genital surgeries⁵, mental health, wellbeing and memory^{6,7}, improved communication⁸ and information sharing through the development of common registries and research and clinical networks^{9,10}. These latter aspects which focus on collaboration and relationship building are critical if partnerships between individuals, communities, professionals, organizational and voluntary groups are to flourish^{11,12}. Here, we report on a workshop to explore the feasibility of using this approach as a platform to allow individuals to inform professional learning as well as provide a space for professionals to ask for feedback around research topic areas.

As part of its intended work plan, the Working Group on Experiences & Perceptions in research COST Action DSDnet¹³, had performed successful e-mail based surveys of specialist centres¹⁴ and professionals providing psychological support¹⁵ to identify variations in practice and the professional needs of these staff. A similar attempt to understand the needs of patients and support group was not successful. Thus, with the help of those from the support group community who were members of the Working Group, a face to face workshop was held.

Method

A sub-set from the working group consisting of five experienced individuals: a parent and support group member (JH, parent and support group lead), nurse (CdS, paediatric), social scientist (CmS, co-opted member), endocrinologist (AK) and psychologist (AD) from three different countries collaborated to plan the workshop. The workshop was described as voluntary, participatory, informal and time limited. It was intended that the population attending the workshop would comprise of some professionals and people with contemporaneous experience of a wide range of conditions that may affect sex development. The following section briefly summarizes the workshop planning and delivery.

Workshop overview and attendees

To determine the likelihood of engagement from individuals with DSD or parents or support group representatives, the working group asked DSDnet members (i.e. endocrinologists, psychologists, urologists, geneticists, nurses and patient representatives) to consider approaching their local support group or patient advocates within their practice to invite them to attend a workshop. Amongst the final 33 attendees, there was a patient and a health care professional from the same centre from 9 different European countries (Austria, Belgium, Bulgaria, Germany, Ireland, Italy, Netherlands, Spain, United Kingdom). In addition, there were 5 professionals who ran the breakout sessions and there were also 10 people who represented support groups. The background of the professionals included endocrinology (9), psychology (2), nursing (1), sociology (1) and urology (1). The age of the patients ranged between 19 and 58 years. All understood English and were comfortable with using an interpreter when necessary. Prior to the workshop all attendees received detailed information about

the event which included reassurance that any information shared outside of the workshop setting would be anonymized.

The workshop schedule included three small group facilitated episodes involving attendees and the professional who had invited them. The four breakout sessions provided opportunity for conversation and discussion about future research topics. The structured small group work explored three key areas; 1) experiences around diagnosis, 2) childhood and young people experiences, and 3) experiences of transition to adult services. Following each of these sessions feedback was shared with the larger group. The four breakout sessions ran concurrently over an extended lunch period and comprised of short presentations followed by discussion. All attendees at the workshop had the opportunity to join at least three of the breakout sessions. The presentations in the breakout sessions focused on the I-DSD registry (SFA), future research areas (AK), obtaining consent in practice (MC, AS), patient education and information resources (NC). The following section highlights the key outputs from the workshop within each of the seven areas discussed.

Results

Experiences around diagnosis

Central to the experience was a need for medical staff to be knowledgeable about such rare condition. Non-expert staff were reported as poor at providing accurate, sensitive and relevant information. Information sharing had the potential to be timely and thoughtful or alternatively burdensome when delivered in a brief time frame. Secrecy around diagnosis and disclosure was, for some individuals, problematic since early experiences influenced confidence in sharing narratives about self (or the child in the case of parents) with others.

However, several individuals reported building trusting relationships with expert professionals. A couple of individuals talked about the experience, or the hope, of having strong partnerships with professionals to talk about bullying, developing resilience, and learning how to safely disclose about self to others. What was raised was the possibility of youth talking with young adults with DSD, perhaps in a mentorship capacity, as a way in which to become better informed. The importance of working with a psychologist was a common thread for parents, and children.

Childhood and young people experiences

During childhood and into adolescence many of the individuals reported challenges to personal body integrity, privacy, and a lack of respect especially linked to requests for physical examinations from medical staff. Absence of conversations with professionals around specific topic areas, such as general life and future family planning options, was reported as a missed opportunity to foster understanding. Many individuals recalled the time spent during childhood at hospital visits as tolerable but significantly burdensome in adolescence.

Most individuals and parents shared the value of their experiences when professionals talked with them in humane ways. As a result of professional continuity both individuals and parents had been able to build strategies that allowed them to share information with others when needed, such as schools thereby maintaining the child's integrity and privacy.

Experience of transition to adult services

For those individuals who had moved to adult care a core concern was the haste and loss of control around transition. This resulted in uncertainty, and for many problems

in communication with new professionals who were less confident to care for them and had little or no knowledge about their condition. For those that had access to correspondence between providers that included incorrect or misleading information this compounded difficulties with building new health relationships. A lack of support networks, and understanding of how to apply information sourced from the internet to self was problematic for a few patients during this phase into early adulthood.

When young adults found connections using social media and support groups there was increased confidence. The need for psychological support into adulthood was highlighted by all the groups since a great deal of learning about self was common in the first decade following transition from paediatric services. A shift in the social discourse around gender was discussed as a way in which natural biological variations could be framed.

I-DSD registry

The rationale for the registry and future development were shared with attendees. A critical discussion focused on the value registries offered, their role in patient safety, and as a platform to build connections. Attendees considered that the registry should cover the life course with the caveat that registrants had the right to withdraw or cancel information at any time. The value of a registry in raising awareness of rare conditions and directing healthcare policy was discussed.

Future research areas

Attendees talked about how research results in some areas of DSD study (i.e., outcome of studies) were not accessible to people with these conditions or families of a child with DSD. Several attendees shared a belief that while a focus on genetic

aetiology/diagnostics in DSD is important it should not be at the expense of other important aspects of care, such as overall wellbeing. The risk of future cancer and the significance of fertility preservation were key topics raised by attendees.

Obtaining consent in practice

Attendees consider the importance of consent in key decision-areas such as hormone research, fertility outcomes and understanding cancer risks. Exploration of gender identity in the clinical setting was believed to be very sensitive and should be done with great caution. Attendees considered that when asking participants to join such a study that researchers be mindful that such requests could be emotive, since it may initially remind people of their differences. In addition, sex and gender studies can raise unnecessary doubts in children, according to both parents and adults, with a few attendees questioning studies where gender identity is usually not an issue, such as CAIS.

Patient education and information resources

For many attendees there was a recollection of brevity linked to oral information, over emphasis on genetic detail or complex language used when talking with professionals. While positivity in early discourses was critical, checking the trustworthiness of information was also important. Several attendees spoke about wanting to have met or connected with adults or self-confident youth with similar rare conditions or connected with support groups. Resource requirement varied but included easy to read materials, well designed and presented, up-to-date information in various formats. Furthermore, opportunity to connect with real life stories where such issues as the ‘inconveniences’ that DSD may bring and unbiased information about surgery can be accessed.

Discussion

Both information sharing and discussion transpired during the workshop between all the attendees. The dynamic of bringing together individuals from various European communities prompted questions and the sharing of information. In addition, including individuals, parents and professionals while focusing the dominant discourse on patient focused issues perhaps resulted in greater collaborative learning for all attendees. Using various workshop engagement approaches, smaller groups and wider discussion allowed attendees to report feeling comfortable which generated discussion across a range of, at times, emotive topics. Workshops that consider the needs of all attendees can be replicated in various contexts, acute care settings, and local communities of practice since there is likely sufficient knowledge, skills, and access to resources within interdisciplinary DSD teams⁴.

Early engagement which is renewed across the life course¹⁶, with experienced and knowledgeable professionals continues to be essential to build trust that can embody the varying emotional needs¹⁷ of individuals with DSD or their parents. When professionals are cognizant of communication approaches, style and language the inherent power dynamic between provider and patient can begin to rebalance, which is especially significant when caring for individuals with these rare conditions and their families¹⁸. The emancipatory lens through which some attendees shared the significance of support groups as a protective factor over the course of their lives was evident in the small group discussion when attendees talked about decision-making, information sharing, and resilience. The complexity around decision-making for this group of patients continues to be discussed in various literature¹¹.

Paying attention to providing sufficient time in dialogues with individuals, sharing up-to-date information in thoughtful, varied, and applicable ways has the capacity to encourage intelligent decision making on the part of parents or professionals¹⁸. Attendees provided insights and offered valuable contributions to future research directions, reflecting and discussing the value and significance of registries as well as expressing the importance of being involved in research topic prioritizing.

Based on evaluating process and summarising findings from our workshop we believe that future workshops could be designed to meet local need. Such workshops can be delivered in meaningful ways, used as a platform to build trust and, foster open communication thereby facilitating learning and promoting education.

Conclusion

Designing, delivering and evaluating workshops locally may be one way in which local DSD interdisciplinary teams, in partnership with support groups and in collaboration with individuals with DSD or their parents, could begin to build networks and communities for those living with rare conditions.

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