

## PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

### ARTICLE DETAILS

<b>TITLE (PROVISIONAL)</b>	Understanding the needs of professionals who provide psychosocial care for children and adults with Disorders of Sex Development.
<b>AUTHORS</b>	Dessens, Arianne; Filho, Guilherme; Kyriakou, Andreas; Bryce, Jillian; Sanders, Caroline; Nordenskjold, Agneta; Rozas, Marta; Iotova, Violeta; Ediat, Annastasia; Juul, Anders; Krawczynski, Maciej; Hiort, Olaf; Ahmed, Faisal

### VERSION 1 - REVIEW

<b>REVIEWER</b>	Wisniewski, Amy Cook Children's Hospital, USA Competing interests: none
<b>REVIEW RETURNED</b>	07-Jun-2017

<b>GENERAL COMMENTS</b>	<p>This is an interesting paper that deserves to be published. However, the following comments should be addressed to strengthen this work even more:</p> <ol style="list-style-type: none"> <li>1. I believe information of psychosocial care of this population has recently been published by David Sandberg; thus, it should not be stated that this is the first report of this information.</li> <li>2. Is it possible to know if results differ if children have ambiguous genitalia</li> <li>3. Is it possible to know if results differ for families who do not pursue surgery for ambiguous genitalia?</li> <li>4. Did participation rates differ between European and non-European countries?</li> <li>5. Were high rates of gender reassignment due to the inability to diagnose the DSD between European and non-European countries?</li> <li>6. Were there differences in gender dissatisfaction between European and non-European countries?</li> </ol>
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<b>REVIEWER</b>	Sandberg, David E. University of Michigan, USA Competing interests: None
<b>REVIEW RETURNED</b>	19-Jun-2017

<b>GENERAL COMMENTS</b>	<p>This manuscript examines a very important, yet relatively neglected topic in the clinical care for persons born with disorders/differences of sex development (DSD): the psychosocial component of multidisciplinary care. The authors conducted an international survey of providers of services from varying disciplines involved in caring for this population. The participation rate was low (n=46; 49%) and participants considered to be eligible for providing psychological services included teachers, religious workers and nurses (in addition to the qualified categories of social work, psychology and psychiatry).</p>
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	<p>Specific Comments</p> <p>Abstract</p> <p>1. I.41: spell out DSD the first time it appears.</p> <p>2. I.51: "Focus psychological interventions included;" change to "The focus of psychological interventions included."</p> <p>3. I. 56: insert "and" prior to "sadness and depression."</p> <p>4. I.55-56: "Psychosocial care provided to parents facilitates acceptance, which is conditional to become affectionate caretakers." This is a strong statement without supporting evidence in the manuscript. One does not need "evidence" to counsel anyone who is distressed; it's simply the right thing to do. However, the claim that this counseling facilitates acceptance, etc requires evidence, but is not provided with data or references in the body of the manuscript.</p> <p>Introduction</p> <p>5. I.64: The correct term is "disorders" of sex development, not "differences." I am familiar with the concerns that some people with these conditions and their advocates have for the term disorder. However, the references to the consensus statement use "disorders of sex development." The authors can certainly express a preference for a different term but, at a minimum, should explain by an alternative term was adopted. It should also be kept in mind that "difference of sex development" is not listed in the ICD.</p> <p>6. I.76: "Some parents prefer to delay gender assignment and/or surgical decision-making, ..." A reference is needed. There is little published evidence that there has been much change in timing of gender assignment or surgery. Further, "delay" without a range may be confusing to readers outside this area: 1 month, 1 year, until the child declares themselves a boy/a girl/neither?</p> <p>7. I. 82: consistency in the use of "gender" vs "sex." I believe the authors are referring to "bodily changes of the other sex"</p> <p>8. I.82-84: please provide supporting references, and similarly for the statements that follow in the paragraph.</p> <p>9. I. 89-94: these generalizations require citations.</p> <p>Results</p> <p>10. Some might strongly contend that it is inappropriate for teachers, religious workers and nurses (I.145-146) to be providing "psychological counseling (e.g., I. 169). I recognize that this statement refers to only a minority (30%) of study participants, but it begs the question of what is considered to be "psychological" or "psychosocial" services in this clinical context.</p> <p>11. I.156: it's unclear what data the chi-square is being used to test</p> <p>12. I.163: "Referrals for psychosocial counseling..." this implies that a medical specialist is the "gatekeeper" to psychosocial services and suggests a model of care other than the multidisciplinary model recommended in references 1 and 2. Do the results of the survey suggest that most psychosocial services are delivered on a consultation basis only? If so, there are presumably many patients/families that do not receive support because there are no obvious signs of distress. Please clarify.</p> <p>13. The description of the services is vague and it is likely that they are delivered in a highly variable manner across providers leaving the reader without a strong sense of what is actually done. For example, I.180-182: Medical providers would likely say that they also deliver some of this counseling. How is that distinguishable from what psychosocial providers deliver?</p> <p>14. Figure 3 and related comments in Discussion: It should be</p>
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	<p>pointed out that the experiences ranked stem from those patient/families “referred” to be seen by the psychosocial provider. Because only a subset of DSD patients/families are referred, this may give the reader a distorted view of the prevalence and pattern of these issues.</p> <p>15. Figure 1. The legend “Most, Middle, Least” is not readily understood.</p> <p>16. Figure 2. “coping and acceptance (remove DSD)”; “Coping and acceptance infertility” to Infertility: coping and acceptance; “Education on psychological aspect DSD” to Education on psychological aspects of DSD.</p> <p>17. Figure 3. “Types of emotional problems seen among individuals (sic) with DSD?” “Types of adjustment problems in DSD”</p> <p>18. for all figures, consider ordering rows from most to least frequent</p> <p>Discussion</p> <p>19. I.230-232: incomplete sentence</p> <p>20. I.236-238: was this information collected as part of the survey or is it assumed?</p> <p>21. Neither the data collected nor Discussion are sufficiently specific to inform the development of psychosocial services tailored to the DSD population. If the approaches to treatment are the same as for other chronic conditions, then that should be stated along with references to evidence-based psychosocial interventions.</p>
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### VERSION 1 – AUTHOR RESPONSE

Reviewer(s)' Comments to Author:

Reviewer: 1

#### Comments to the Author

This is an interesting paper that deserves to be published. However, the following comments should be addressed to strengthen this work even more:

1. I believe information of psychosocial care of this population has recently been published by David Sandberg; thus, it should not be stated that this is the first report of this information.

Response: We adapted the manuscript lines 244-248 and referred to Rolston’s publication (published June 2017, after submission of our manuscript)

2. Is it possible to know if results differ if children have ambiguous genitalia

3. Is it possible to know if results differ for families who do not pursue surgery for ambiguous genitalia?

Response: We fully agree that these are interesting questions but unfortunately we are not able to answer based on our available data. Focus of the survey was activities of professions who provide psychosocial care in patients with DSD and their parents.

4. Did participation rates differ between European and non-European countries?

Response: This is a relevant comment. A Providers of psychosocial care in DSD are not well organised (there is no professional organisation). We were able to identify email addresses of providers of psychosocial care from multiple sources related to DSD as described in the Methods section lines 278 - 282. From the email addresses it is difficult to trace location, hospital or institute. Many potential responders had gmail and hotmail accounts. So we only know location, hospital and institute names from participants.

5. Were high rates of gender reassignment due to the inability to diagnose the DSD between European and non-European countries?

6. Were there differences in gender dissatisfaction between European and non-European countries?

Response: Unfortunately, we cannot detail this relevant issue. Focus of the survey was activities of professions who provide psychosocial care. As such we did not investigate gender (re)assignment, gender dissatisfaction etc. Such a study demands another study design.

Reviewer: 2

Comments to the Author

This manuscript examines a very important, yet relatively neglected topic in the clinical care for persons born with disorders/differences of sex development (DSD): the psychosocial component of multidisciplinary care. The authors conducted an international survey of providers of services from varying disciplines involved in caring for this population. The participation rate was low (n=46; 49%) and participants considered to be eligible for providing psychological services included teachers, religious workers and nurses (in addition to the qualified categories of social work, psychology and psychiatry).

In the U.S., my experience has taught me that institutional review boards would commonly provide a waiver for a survey of this sort. However, I don't know what the practices are in Europe. I see no mention in the paper regarding how this issue was addressed. Also, it is not stated whether respondents were promised anonymity? confidentiality? Was their informed consent?

Response: See lines 155-157. All potential respondents received a personalized email with an introduction to the present survey, in which they were informed about the relationship to COST-Action BM1303 (with reference link) and the purpose of the survey. By clicking the reference link to the survey, respondents gave their informed consent. We asked respondents to fill out their contact details so participation was not anonymous. But respondents could choose to skip questions, if they felt uncomfortable to answer.

Specific Comments

Abstract

1. l.41: spell out DSD the first time it appears.

Response: We adapted the manuscript

2. I.51: "Focus psychological interventions included;" change to "The focus of psychological interventions included:"

Response: We adapted the sentence

3. I. 56: insert "and" prior to "sadness and depression."

Response: We adapted the sentence

4. I.55-56: "Psychosocial care provided to parents facilitates acceptance, which is conditional to become affectionate caretakers." This is a strong statement without supporting evidence in the manuscript. One does not need "evidence" to counsel anyone who is distressed; it's simply the right thing to do. However, the claim that this counseling facilitates acceptance, etc. requires evidence, but is not provided with data or references in the body of the manuscript.

Response: We adapted the sentence and added: It's assumed that parental support ...

#### Introduction

5. I.64: The correct term is "disorders" of sex development, not "differences." I am familiar with the concerns that some people with these conditions and their advocates have for the term disorder. However, the references to the consensus statement use "disorders of sex development." The authors can certainly express a preference for a different term but, at a minimum, should explain by an alternative term was adopted. It should also be kept in mind that "difference of sex development" is not listed in the ICD.

Response: We agree with this comment on nomenclature. We adapted the manuscript accordingly.

6. I.76: "Some parents prefer to delay gender assignment and/or surgical decision-making, ..." A reference is needed. There is little published evidence that there has been much change in timing of gender assignment or surgery. Further, "delay" without a range may be confusing to readers outside this area: 1 month, 1 year, until the child declares themselves a boy/a girl/neither?

Response: The reviewer is right. In this paragraph however, we paint the picture of the neonatal period, which can be extremely distressing for most parents (see reference 3) and in which psychosocial consultation is often required.

7. I. 82: consistency in the use of "gender" vs "sex." I believe the authors are referring to "bodily changes of the other sex"

Response: The reviewer is right. We adapted the text accordingly.

8. I.82-84: please provide supporting references, and similarly for the statements that follow in the paragraph.

Response: We added supporting references 4-8

9. I. 89-94: these generalizations require citations.

Response: Thank you for this valid suggestion. We added supporting references 10-18

## Results

10. Some might strongly contend that it is inappropriate for teachers, religious workers and nurses (I.145-146) to be providing “psychological counseling (e.g., I. 169). I recognize that this statement refers to only a minority (30%) of study participants, but it begs the question of what is considered to be “psychological” or “psychosocial” services in this clinical context.

Response: We agree with the reviewer. We adapted the manuscript.

11. I.156: it's unclear what data the chi-square is being used to test

Response: Thank for you pointing to this lack of information. We have now added missing information

12. I.163: “Referrals for psychosocial counseling....” this implies that a medical specialist is the “gatekeeper” to psychosocial services and suggests a model of care other than the multidisciplinary model recommended in references 1 and 2.

Response: We asked responders how patients and parents were referred to their services. Self-referral was one of the options that could be indicated, but this option was rarely selected. The survey conducted by Kyriakou et al. (ref 19) revealed that for the large majority of DSD patients, the pediatric endocrinologist is considered the key medical specialist in the diagnostic process, in clinical management and follow-up until adulthood. This key position makes it likely that parents will discuss their difficulties in coping first with their pediatric endocrinologist, as he/she is best known. Knowing parents and their children well, the pediatric endocrinologist may be the first professional to observe patient's and parents needs for psychosocial consultation.

Do the results of the survey suggest that most psychosocial services are delivered on a consultation basis only? If so, there are presumably many patients/families that do not receive support because there are no obvious signs of distress. Please clarify.

Response: From our findings we cannot draw the conclusion that most psychosocial services are delivered on a consultation basis only. We did not conduct a survey among patients/families, so we cannot answer questions regarding psychosocial services hoped for but not received. We could not compare responses from our responders with data from other resources such as information from hospital registrations, registries like i-DSD or epidemiological databases.

13. The description of the services is vague and it is likely that they are delivered in a highly variable manner across providers leaving the reader without a strong sense of what is actually done. For example, I.180-182: Medical providers would likely say that they also deliver some of this counseling. How is that distinguishable from what psychosocial providers deliver?

Response: the reviewer is right. In this survey we aimed to investigate professional training and activities of professionals in psychosocial care worldwide. As local organization of health care and the services of providers of psychosocial care differ between nations, our survey only focused on services expected to be delivered by the majority of colleagues. We did not carry out a survey on psychosocial care provided by medically trained personnel so we cannot compare psychosocial care delivered by medically trained professionals and professionals in psychosocial care.

14. Figure 3 and related comments in Discussion: It should be pointed out that the experiences ranked stem from those patient/families "referred" to be seen by the psychosocial provider. Because only a subset of DSD patients/families are referred, this may give the reader a distorted view of the prevalence and pattern of these issues.

Response: this survey was designed to obtain an overview of delivered services by providers of psychosocial care from different nations. It was not our intention to assess prevalence and patterns.

15. Figure 1. The legend "Most, Middle, Least" is not readily understood.

16. Figure 2. "coping and acceptance (remove DSD)"; "Coping and acceptance infertility" to Infertility: coping and acceptance; "Education on psychological aspect DSD" to Education on psychological aspects of DSD. 17. Figure 3. "Types of emotional problems seen among individuals (sic) with DSD?" "Types of adjustment problems in DSD"

18. for all figures, consider ordering rows from most to least frequent

Response: the reviewer attentively read the figures. We adapted the figures in line with these suggestions, but did not order rows from most to least frequent as this requires to make all figures over again. The bars in the figures do not reflect an ordinal principle.

## Discussion

19. I.230-232: incomplete sentence

Response: The reviewer is right. We adapted the manuscript.

20. I.236-238: was this information collected as part of the survey or is it assumed?

Response: see figure 1, information collected in the survey.

21. Neither the data collected nor Discussion are sufficiently specific to inform the development of psychosocial services tailored to the DSD population. If the approaches to treatment are the same as for other chronic conditions, then that should be stated along with references to evidence-based psychosocial interventions.

Response: We agree with the reviewer. This survey was only designed to assess services delivered by providers of psychosocial care for patients/parents with DSD.. The survey was not intended to assess standards of care for patients, but findings in this survey may be helpful in development of standards of care.



## Correction: *Understanding the needs of professionals who provide psychosocial care for children and adults with disorders of sex development*

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Dessens A, Guaragna-Filho G, Kyriakou A, *et al.* Understanding the needs of professionals who provide psychosocial care for children and adults with disorders of sex development *BMJ Paediatrics Open* 2017;1:e000132. doi: 10.1136/bmjpo-2017-000132.

The funding information for this paper was missed off by the production team, we would like to apologise for this oversight. This survey was conducted as part of the COST Action BM 1303 DSDnet, supported by COST (European Cooperation in Science and Technology, [www.cost.eu/COST\\_Actions/bmbs/BM1303](http://www.cost.eu/COST_Actions/bmbs/BM1303)). The International Disorder of Sex Development (I-DSD) Registry is supported by Medical Research Council partnership award G1100236 (Ahmed, Bryce, Jiang, Lucas-Herald, Rodie and Sinnott) and was initially developed under a project grant from the Seventh European Union Framework Program (201444) and a project grant from the Research Unit of the European Society for Paediatric Endocrinology. Guaragna-Filho was supported by CAPES Foundation, Ministry of Education of Brazil (BEX 3547–15–9).

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