

Meeting between experts: evaluation of the first UK forum for lay and professional experts in intersex

Sarah M. Creighton^{a,*}, Catherine L. Minto^a, Lih Mei Liao^b,
Julie Alderson^c, Margaret Simmonds^d

^a Department of Obstetrics and Gynaecology, Elizabeth Garrett Anderson Hospital, University College Hospitals,
Huntley Street, London WC1 6DH, UK

^b Sub-Department of Clinical Health Psychology, University College London, London, UK

^c Department of Health Psychology, Leeds Teaching Hospitals, Leeds, UK

^d Androgen Insensitivity Syndrome Support Group (AISSG),¹ P.O. Box 269, Banbury, UK

Received 13 October 2002; received in revised form 4 May 2003; accepted 9 June 2003

Abstract

At present the clinical management of intersex is in turmoil. The policy of non-disclosure of diagnosis is widespread and cosmetic genital surgery is routinely performed on infants throughout the world. Some clinicians feel such practices are in the interest of the intersex child and the family, but some intersex adults are calling for a moratorium on sex assignment genital surgery. These widely opposing views have led to distrust between groups. One way to begin to address these critical issues is to facilitate dialogue with equal input from clinicians and intersex people and families. Clinicians are experts by training, but patients and families are experts through lived experiences. Our paper reports the rationale, process and outcome of the first UK forum bringing together these different experts to address some of the most complex issues in clinical services. In communicating our experiences, we hope that it will provide a useful reference point for those seeking similar service-user/provider collaboration in other areas of medicine.

© 2003 Elsevier Ireland Ltd. All rights reserved.

Keywords: Intersex; Multidisciplinary; Genital surgery; Psychology; Expert patient

1. Introduction

“The Expert Patient” document, published in August 2001, represents a new approach to chronic disease management for the 21st century [1]. It sets out proposals to allow patients to play a central role in disease management. Although the report focuses on common chronic illnesses such as asthma, diabetes and arthritis, the concept of the expert patient is perhaps even more important for those with rarer conditions such as the so-called “intersex” conditions. A patient with an intersex condition may be the only case that their doctor ever comes across. The rarity of such conditions increases a sense of isolation and powerlessness.

In intersex conditions the dual pathways of sexual determination and differentiation leading to a male or female child do not proceed as expected. This leads to the birth

of a child with internal and/or external physical characteristics of both sexes. These conditions are rare and include for example a baby born with ambiguous genitalia, a girl with primary amenorrhoea found to have an XY karyotype, or a man undergoing infertility investigations and found to have an XX karyotype. Current controversies in intersex management are centred on the disclosure of karyotype and the role of infant sex assignment genital surgery [2].

If an intersex child is assigned to a female sex of rearing, current practice is to perform ‘feminising’ genital surgery in the first year of life [3]. This usually involves clitoral reduction and vaginoplasty. It has been widely assumed that that surgery would lead to a more stable gender identity and greater psychological well being [4]. Such assumptions have been strongly criticised on conceptual grounds [5–7]. Furthermore, there has never been any reliable empirical evidence linking genital surgery to better outcomes. If anything patient forums have documented severe psychological distress amongst some of their members [8–10]. These accounts are corroborated by recent psychological analyses [11,12]. Affected adults have been increasingly vocal about their

* Corresponding author. Fax: +44-20-7380-9565.

E-mail address: sarah.creighton@uclh.org (S.M. Creighton).

¹ An international peer support group for adults and families affected by androgen insensitivity syndrome (<http://www.medhelp.org/www/ais>).

dissatisfaction with clinical decisions made on their behalf during childhood or adolescence. It has not been possible to ascertain what proportion of patients regret their surgery, but surgical sex assignment for intersex is also increasingly criticised on ethical ground [13].

The uncertainty brought about by a lack of reliable longitudinal data leaves considerable scope for disharmony between doctors and patients leading in a few cases to litigation. In the light of such a climate, service-user involvement is crucial for improving working relationships. Progress cannot take place in the absence of constructive dialogue. This was the rationale for the open forum and debate reported in the current article. Given the potential for antagonism between service providers and users however, a meeting between lay and professional experts would always raise concerns and this may be why it had not taken place in the context of intersex services. The rest of this article reports the process and outcome of the first attempt in the UK to bring about such a meeting.

2. The forum: funding and organisation

The forum has been conceptualised and developed jointly by members of the Androgen Insensitivity Syndrome Support Group (AISSG) and clinicians from University College London Hospitals and the Leeds Teaching Hospitals, both of which run multidisciplinary intersex clinics. The AISSG is a patient peer support group that came into existence in 1988. It currently has 110 UK parent/adult subscribers and an enquiry list of several hundreds. It has an extensive web site (<http://www.medhelp.org/www/ais>) and a regular biannual newsletter 'ALIAS'.

AISSG has spent some years liaising with clinicians and has long been keen to have a formal meeting with interested clinicians. Initial discussion at a national AISSG meeting identified themes that members wished to discuss. It was agreed that a multidisciplinary forum was needed. An organising committee composed of clinicians and AISSG members was set up and a draft programme agreed. A small charge had to be made for the meeting, however financial assistance was made to those who required it. All speakers waived travel expenses and no honorariums were paid.

The meeting was advertised by e-mail and post to all members of the AISSG. It was also advertised in the newsletter of the British Society for Paediatric and Adolescent Gynaecology (BritSPAG), which has over 200 members from varied clinical specialities and is likely to reach many of those interested in this area of medicine. Other clinicians and researchers known to be active in this area were also sent details of the meeting. The meeting registration form asked delegates to specify, if they wished, whether they were a health professional (including area of speciality), patient/consumer, parent or relative of an intersex patient, or other interested party.

2.1. The forum

The aim of the meeting was to enable clinicians, patients and parents to engage in open dialogue on equal footing. The programme focused on the controversial areas in intersex including disclosure and surgery. Speakers comprised clinicians actively engaged in practice and research and leading members of the support group. All sessions were jointly chaired by a clinician and support group representative. The programme also included a video of the personal experiences of two women who had undergone treatment for intersex. The final session provided an opportunity to begin to formulate recommendations for future intersex management.

2.2. Evaluation

A formal feedback questionnaire was designed to evaluate the day. This was adapted from a standard evaluation assessment used by one of the authors (LML). Completion of the questionnaire was anonymous but delegates were asked to indicate if they were a professional (including area of speciality), patient/consumer, parent of an intersex patient, or other interested party. The questionnaire was divided into two sections: the first evaluated overall delegate satisfaction and dissatisfaction and the second section focused on feedback of specific presentations.

Two of the questions involved completion of a linear analogue scale about the satisfaction felt by each delegate:

How satisfied do you feel about today?

To what extent were you able to express what you had wanted to express?

The score ranged from 1 for "not at all" to 5 for "very much so". Statistical analyses of satisfaction scores comparing the patient and clinician groups, and different clinical speciality groups, were performed with SSPS software using Pearson chi-squared test.

There were also four open questions to determine key issues which may contribute to the delegates feeling of satisfaction or dissatisfaction with the meeting:

One thing about today you feel most satisfied with?

One thing about today you feel least satisfied with?

How did you feel whilst discussing and thinking about intersex in this forum?

What, if anything, would have helped?

Responses to these questions were content-analysed by an independent observer who had not been involved in the event. A total of 29 response categories were generated for the responses to the open questions. Examples of the categories included a process for sharing, learning important information, the multidisciplinary approach, inclusion of user groups, contact with specific clinicians and increased awareness. Each returned questionnaire was coded for the

presence or absence of each of the 29 categories generated by the independent observer. An inter-rater reliability check was carried out for a sub-sample of the questionnaires by two of the authors (LML and SMC) who were blinded to the independent observer’s and each other’s coding. Agreement was high ranging between 83 and 100% with a mean of 93%.

3. Results

One hundred and forty-eight delegates registered for the meeting and 140 attended on the day (Table 1). The delegates comprised of fewer patients and relatives (30%) than healthcare professionals (65%). There was a good spread of healthcare professionals from the various involved clinical areas.

Eighty-six (61%) questionnaires were returned. These were evenly distributed between the different delegate cat-

Table 1
Delegate details from registration form

Delegates categories	Number
Patients	22
Parents or relatives	20
Health professionals	91
Endocrinologists and paediatricians	26
Gynaecologists	21
Surgeons and urologists	18
Psychologists and psychology researchers	14
Specialist nurses	12
Other or unknown	7
Total	140

egories. The majority of delegates answered the question on satisfaction (85/86). Fewer delegates (71/86) answered the question regarding their ability to express, and all of those who did not respond to this question were healthcare professionals.

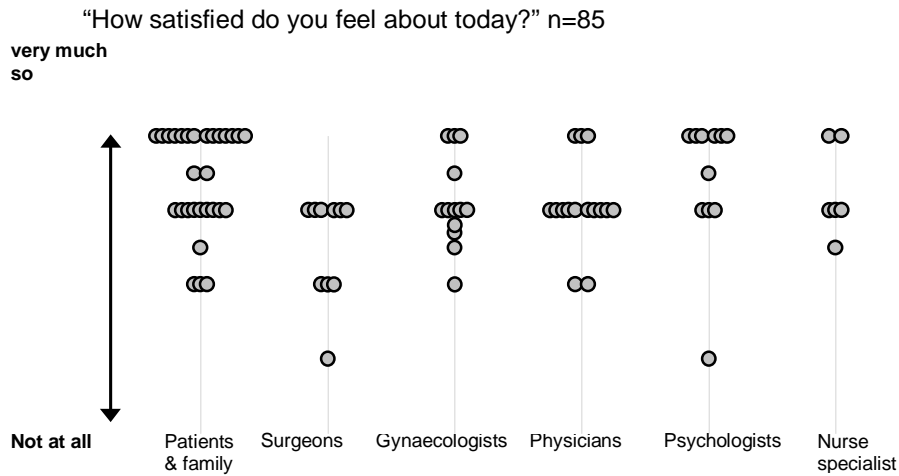


Fig. 1. Evaluation analysis of Question 1. “How satisfied do you feel about today?” (n = 85).

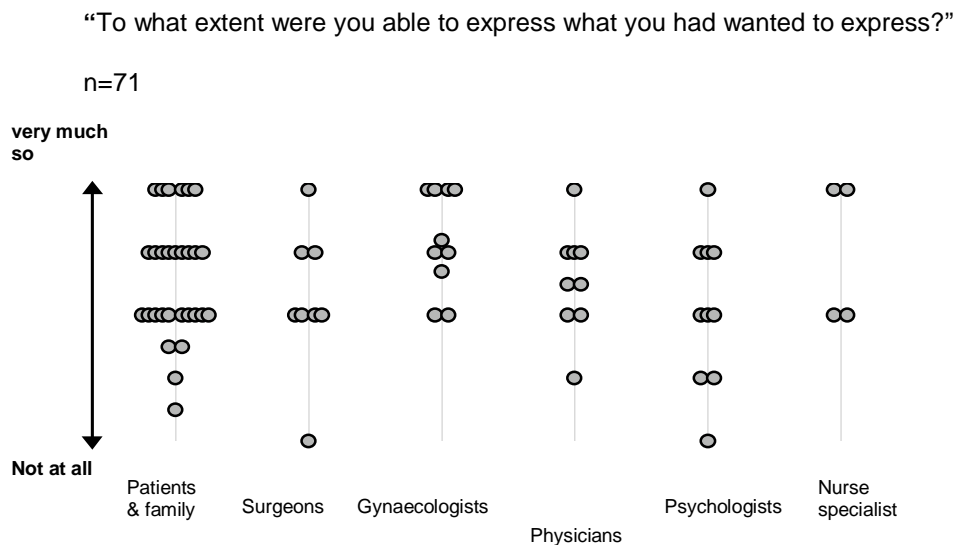


Fig. 2. Evaluation analysis of Question 2. “To what extent were you able to express what you had wanted to express?” (n = 71).

Table 2
Themes from analysis of open questions

	Most common theme			
	One thing about today you feel most satisfied with?	One thing about today you feel least satisfied with?	How did you feel whilst discussing and thinking about intersex in this forum?	What, if anything would have helped?
Overall	Sharing environment (e.g. communication, open discussion positive atmosphere) 43%	Not enough discussion time 32%	Positive feelings (e.g. felt comfortable, stimulated, proud, lucky, humbled) 42%	Improvement to content (e.g. parent speaker) 16%, improvement to the information (e.g. more evidence base) 16%
Clinicians	As above 43%	Quality of information (e.g. lack of evidence or research) 40%	As above 34%	Improvement to content (e.g. parent speaker) 23%
Patients/parent group	As above 45%	Not enough discussion time 28%	As above 55%	More time 21%

Overall there were no significant differences in scores for either satisfaction or ability to express when comparing the overall delegate categories of all health professionals compared with patients/parents. However on dividing the healthcare professionals into groups according to speciality (Fig. 1), there were significant differences with surgeons being less satisfied than both the patient/parents group ($\chi^2 = 11.47$, $P = 0.009$) and the psychologists group ($\chi^2 = 10.98$, $P = 0.01$). There were no significant differences in the ability to express (Fig. 2).

Analysis of the open questions identified common themes. Table 2 gives the most frequently given answer in each section. On further inspection of the groups there were some other interesting findings. For example nine (20%) of clinicians (all surgeons or gynaecologists) felt the atmosphere was antagonistic whereas only one patient expressed this. Both groups (28% of professionals and 20% of patients) reported negative feelings such as being upset, angry, sad or exasperated during the forum.

All of the presentations were scored for relevance and content ranging from 1 (minimum) to 5 (maximum). All presentations scored highly and there was no difference between clinician and patient groups.

4. Discussion and conclusion

By far the most salient observation on the day was that both clinicians and patients greatly valued the unique opportunity of the symposium. This was also apparent in the feedback forms. The most frequent response from both professional and lay groups was appreciation of the opportunity for sharing and for open exchanges between diverse groups of people. These processes appeared to have been more valuable than the gaining of new knowledge or clarity about future directions, although these were also expressed in the feedback.

Patient satisfaction was high and the appreciation of the presence of clinicians and of the frank and open discussion was indicated by feedback such as “collaboration of

expertise” and “levelled the playing field”. Some patients also expressed appreciation of doctors’ difficulties.

Overall satisfaction with the meeting was also high for the professional groups. Surgeons were significantly less satisfied and the reasons included “anti-surgical/anti-doctor bias” and feeling “under siege as a surgeon”. Surgeons and gynaecologists were more likely than other clinicians to have experienced the forum as a hostile environment. The forum was potentially an unusual experience for everyone but perhaps particularly for the surgical group, some of whom appeared to have personalised patient criticisms. However, the cost to patients who exposed aspects of their personal lives before a powerful audience in order to offer us insight must not be under-estimated.

Some clinicians expressed intolerance at the lack of ‘objectivity’ expressed by patients as indicated by feedback such as “support group not evidence-based” or “more evidence, less anecdote”. Clinicians and academics claim their expertise through formal knowledge built on empirical evidence, but patients can also claim some expertise through intimate lived experiences [14]. Both accounts can claim the status of ‘evidence’, and both are valuable. If anything, there is rather more information on the long-term trajectory of childhood treatments from patients than from any rigorous empirical research.

When assessing how delegates felt while discussing intersex in an open forum, the most common theme in both groups was of positive emotions, although both groups also expressed negative emotions, e.g. “sad”, “angry”. It is interesting that this question appeared to have been interpreted somewhat differently by lay and professional delegates. Patients were more likely to interpret this question as it was intended—as an enquiry of their emotional state and concerns during discussion of such a complex and troubled subject in a mixed audience. Their replies included “positive and encouraged”, “overwhelmed” and “a sense of freedom and relief”. Many clinicians, on the other hand, interpreted this as an enquiry as to the quality of the meeting with answers such as “good meeting” “very useful”, “very interesting”. Far fewer clinicians commented on

their feelings or more private thoughts; the few that did were more reserved offering feelings such as “encouraged”, “comfortable”, “interested”. This in course reflects the fact that for clinicians intersex is a non-personal subject whilst for patients it is very personal indeed.

4.1. Conclusion

The lessons learnt from this project are relevant not just for intersex experts but also other areas of clinical practice, e.g. obstetrics, assisted reproduction, oncology, genetic screening, sexual health. Despite different thoughts and feelings that circulated in the meeting and varying levels of participation, lay and professional delegates uniformly expressed that the opportunity for open discussion—even on the most emotive topics—was of paramount importance.

We recognise that important subjects such as a moratorium of childhood genital surgery will not be resolved in a meeting such as this and this had not been the intended outcome. However we had hoped to begin an open dialogue that would continue, to remove some of the obstacles in communication, to dispel the notion that people with diverse perspectives are not able to engage in discussion, and to promote a willingness to consider the issues collaboratively despite anticipated differences.

4.2. Practice implications

At present there is little reliable short- or long-term outcome data on any aspect of intersex management. Long-term outcome data relating to sexuality and psychological well being can only be provided by adults. This unique opportunity for clinicians who work with intersex children and their families to meet intersex adults will hopefully pave the way for increasing appreciation of the potential long-term sequelae. Whilst even implied criticism would be disconcerting for clinicians who feel that they have done their best under difficult circumstances, the feedback does remove the scope for complacency and engage us in collaborative problem solving. At the same time, explanations about practices put forward by healthcare professionals could help clarify some of the confusion amongst service users and further the debate amongst themselves. A mutual recognition of each other’s differing knowledge and experience, and a willingness to contemplate change, is critical for progress. We are optimistic that given the positive response by all parties to the idea of dialogue between professional and lay groups, future opportunities for communication are more likely to be taken up, in intersex and other clinical services.

Acknowledgements

The authors would like to thank all members of AISSG who contributed to the meeting, especially Lesley for her tireless fund raising efforts. Thanks are also due to other clinicians involved in organising the meeting including Gerard Conway (UCLH) and Adam Balen (Leeds). We are grateful to Carla Croft for her independent analysis of the questionnaires. Out of the many pharmaceutical companies approached, Novo-Nordisk was the only one interested in the concept and donated £1000 towards the event. A successful application to the Kings Fund project stimulus grants raised a further £1000. Finally the Portland Hospital for Women and Children generously donated £500.

References

- [1] Department of Health The Expert Patient: A new approach to chronic disease management for the 21st century. Available from <http://www.doh.gov.uk/healthinequalities> [August 2001].
- [2] Creighton S, Minto C. Managing intersex. *Br Med J* 2001;323: 1264–5.
- [3] Lawson Wilkins Pediatric Endocrine Society and The European Society for Pediatric Endocrinology. Consensus statement on 21-hydroxylase deficiency. *J Clin Endocrinol Metab* 2002;78:4048–54.
- [4] Money J, Hampson JG, Hampson JL. Hermaphroditism: recommendations concerning assignment of sex change of sex, and psychological management. *Bull Johns Hopkins Hosp* 1955;97:284–300.
- [5] Kessler SJ. *Lessons from the intersexed*. New Brunswick (NJ): Rutgers University Press; 1998.
- [6] Kitzinger C. Intersex and the psychology of women. In: Liao LM, Boyle M, editors. *Intersex [special issue]*. *Psychologist* [in press].
- [7] Morland I. The organ’s apprehension. In: Liao LM, Boyle M, editors. *Intersex [special issue]*. *Psychologist* [in press].
- [8] Androgen Insensitivity Syndrome Support Group UK. Truth disclosure discussion, vol. 1. ALIAS Newsletter from the Androgen Insensitivity Syndrome Support Group; 1998. p. 9–12. Available from <http://www.medhelp.org/www/ais>.
- [9] Androgen Insensitivity Syndrome Support Group UK. Desperation at the doc’s, vol. 1. ALIAS Newsletter from the Androgen Insensitivity Syndrome Support Group; 1999. p. 9–13. Available from <http://www.medhelp.org/www/ais>.
- [10] Androgen Insensitivity Syndrome Support Group UK. Managing your medics. ALIAS 2000;1:255–7. Available from <http://www.medhelp.org/www/ais>.
- [11] Alderson J, Madill A, Balen A. Fear of devaluation: understanding the experience of intersexed women with androgen insensitivity syndrome. *Br J Health Psychol* [in press].
- [12] Liao LM. Learning to assist women born with atypical genitalia: journey through ignorance, taboo and dilemmas. *J Reprod Infant Psychol* [in press].
- [13] D A. Ambiguous sex or ambivalent medicine? Ethical issues in the treatment of intersexuality. *Hastings Cent Rep* 1998;28(3):24–35.
- [14] Frank A. *At the will of the body: reflections on illness*. Boston: Houghton Mifflin; 1991.