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Psychosexual development management of bladder exstrophy epispadias in complex patients

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Summary

Introduction
Bladder-exstrophy-epispadias complex (BEEC) represents a spectrum of urogenital step-wise malformations: epispadias, complete exstrophy, and cloacal exstrophy. Psychosexual development in adolescent patients with BEEC may become especially problematic. At present, there are few contributions in the literature investigating the validity of psychosexual treatment in order to tackle this particularly emotional and personal development phase.

Objective
The study aimed at verifying the efficacy of an intervention methodology for psychosexual support of a group of adolescents with BEEC. The main goal of the intervention program was to educate the adolescents and re-frame how they see themselves or feel about themselves, especially in relation to BEEC. In particular it was predicted that the program could: (1) improve the perception of pleasure concerning the body, particularly regarding the genital area, giving proper and specific information on pleasure, masturbation and medical history of BEEC; and (2) elicit a more relational-affective perspective on sexuality.

Study design
13 adolescent patients took part in the 1-year program. The effects of the intervention program were verified through a test-retest methodology using Sexuality Evaluation Schedule Assessment Monitoring (SESAMO).

Results
The results showed that participants changed their attitude in several psychosexual areas, more specifically: psycho-environmental situations, body experience, areas of pleasure, medical and sexual history, and motivation and conflict areas (Summary Table).

Discussion
This study demonstrated, for the first time, that a targeted program may significantly improve the psycho-sexual condition of adolescents with BEEC. In particular, this research showed that adolescents need to be able to discuss and tackle topics of a psychological and sexual nature, as well as receive understandable answers that can be put into practice in their everyday lives. The study had several methodological limitations, especially owing to the limited number of participants, the absence of a follow-up period of a few months after the intervention, and the overall exploratory nature of the program.

Conclusion
This intervention methodology may be considered a first attempt at improving the self-esteem of adolescents with BEEC, by contrasting forms of psychological difficulties in order to improve the quality of life of these young people.

Table

Areas	Median percentile rank-pre pre-intervention		Median percentile rank-post postintervention	
	M	F	M	F
Related psycho-environmental situations	84*	89*	37	37
Body experience	98*	98*	46	38
Areas of pleasure	90*	90*	37	37
Medical and sexual history	99*	98*	40	40
Motivation and conflict	95*	95*	31	31

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Introduction

Bladder-exstrophy-epispadias complex (BEEC) represents a spectrum of urogenital malformations ranging from epispadias to classical bladder exstrophy and exstrophy of the cloaca. Being the mildest form, isolated epispadia is relatively rare, and occurs with an incidence of 1:117,000 in males and 1:484,000 in females. Incidence of complete bladder exstrophy is about 1:30,000 live births, boys being affected three to four times more frequently than girls. The extreme form BEEC (i.e. cloacal exstrophy) is reported to have an incidence of 1:200,000 to 1:400,000; the ratio of males to females is about 2:1 [1]. In addition to the complex surgical procedure required to correct the severe bladder abnormalities and genital deformities involved in BBEC, other problems may arise in the patient concerning renal function, urinary and faecal incontinence, fertility, and psychosocial and psychosexual development. Adolescents with BEEC have significant physical and mental difficulties due to their genitourinary deformities and, in some cases, faecal incontinence, and the negative impact the two have on mental health and psychological functioning [2].

The high risk of psychiatric disorders amongst these individuals is a consequence of both their suffering from a chronic physical disorder and the characteristics of the actual abnormality [3]. In late adolescence, when relationships with the opposite sex become particularly important [4], males seem to be more affected by this issue than females. The former have less intimate relationships than the latter, perhaps because they experience greater difficulties in dealing with issues regarding sexual activities and the genital system [5], as well as anxiety regarding physical abnormalities (their penis size, its deformed shape when erect [6]), and, when older, it becomes difficult for the opposite sex to accept unnatural methods of tackling incontinence [7]. Patients with BEEC often find it difficult to recognise their own bodies, do not feel like men/women and do not accept themselves. This can lead to deep depression, sometimes resulting in extreme acts such as suicide [8]. However, there are studies providing evidence of effective coping strategies and the capability of achieving good quality of life [9–11].

However, it is believed that no study, to date, has proposed and/or validated an evidence-based methodology with which to support individuals with BEEC during critical stages of their development, such as adolescence. For this reason, the aim of this pilot study was to verify the efficacy of a targeted intervention methodology for adolescent patients affected by BEEC, to help them deal with psychosexual issues. The main goal of the study was to educate the patients and re-frame how they see themselves or feel about themselves, especially in relation to BEEC. In particular, it was predicted that the program could: (1) improve the perception of pleasure concerning the body, particularly regarding the genital area, giving proper and specific information on pleasure, masturbation and medical history of the BEEC; and (2) to elicit a more relational-affective perspective on sexuality.

Materials and methods

Between 2013 and 2014, the families of adolescents that were receiving treatment at the Department of Pediatric Urologic Surgery of the Institute for Maternal and Child Health – IRCCS 'Burlo Garofolo', Trieste, Italy, and who (1) were diagnosed with BEEC, (2) were aged between 14 and 16 years old, (3) had achieved urinary continence, and (4) underwent genital reconstruction interventions were contacted to participate in the present study. Of the 35 contacted families, 26 agreed to participate in the pilot study. Those who refused, did so mainly because of logistics (i.e. difficulty in reaching the hospital where the meetings were organised).

The group comprised 13 adolescents, of whom nine were males (mean age 15, SD 0.60, range 14–16) and four were females (mean age 14, SD 0.60, range 14–16). It was decided to include adolescents aged 14–16 years old because (1) students are introduced to a more exhaustive program on psychosexual education in schools, at least in Italy where the study was carried out, but teachers are rarely able to effectively explain this topic to adolescents with BEEC; and (2) from experience, at that age, both parents and adolescents seem to be better motivated to participate in a program like the one in this study. All adolescents in the sample were born with complete exstrophy (epispadias and exstrophy). As far as participants' relationship status was concerned, with the exception of two adolescents who had never even kissed anybody, all the participants had at least once experienced touching a partner, but they said they had never allowed the partner to touch them, nor had they ever experienced complete sexual intercourse. During their participation in the program, none of the participants were in a stable relationship; two adolescents mentioned having had a short relationship during the summer, which only lasted a couple of months.

The research received a positive review from the Ethics Committee of the Institute for Maternal and Child Health—IRCCS 'Burlo Garofolo', Trieste, Italy.

The study schedule included 12 meetings. The first meeting was with the paediatric urology surgeon in the presence of a psychologist with specific training in urogenital and sexual disorders. The remaining meetings were with the same psychologist only. Each lasted approximately 60 min and occurred on a monthly basis, for 12 months, in a quiet room at the Institute.

To evaluate the effects of the intervention, Sexuality Evaluation Schedule Assessment Monitoring (SESAMO) was used. This evaluation test was created in Italy, validated and standardised with reference to the characteristics of the Italian population, according to ISTAT data relating to the 1991 census. This test was chosen because it can detect potential physical and psychological issues linked to the psychosexual-relationship sphere [13,14], in particular: relevant factors that were still under-investigated in relation to BEEC adolescents [15,16].

The selected SESAMO version was used in its standard clinical form: the test was divided into four sub-groups, based on the sex and the relationship status at the moment of entry into the program; groups were divided on the basis

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of the participants' sex, and, within each group, a further division was operated on the basis of the participants' involvement in a relationship — or lack of it. The questionnaire was composed of 62 items for single males and 81 items for males in a relationship, while there were 64 and 85 items for single and non-single females, respectively [13,14]. The SESAMO is known to have also been used in international contexts [17—19]. The test was in 'paper and pencil' form and answers were analysed using conversion grills both in z-scores and in percentile ranks for each area. The rough scores of each participant were standardised. The SD and the mean were calculated for each item using SESAMO-Win (Microsoft, USA). These values were used to establish the z-scores and the percentile ranks in order to assess the gaps between the scores of the subjects for every single item. This final data allowed the unease of the subjects (or lack thereof) to be assessed on the basis of the values' positioning above or below the z-score equal to 0 and/or the 50th percentile. The areas that presented a positive z-score, or that ranked above the 50th percentile, were areas to be further examined as problematic [20].

Participants were evaluated 1 month prior to access (within a range of 25—34 days) and 1 month after completion of (within a range of 24—35 days) the program. Questions and remarks spontaneously arose during the training and the psychologist, also on the basis of previous clinical experience [21], tried to answer them during the program (Table 1).

Intervention description

The intervention methodology used a cognitive approach to provide a framework of significance for BEEC and show the significance it has for these patients. More specifically, the covered topics included:

(1) Providing information on what BEEC is, to (a) provide a coherent and univocal message and (b) increase trust and respect for the operators, who had equal levels of importance and skill despite their different professional disciplines.
(2) Verbalising emotions linked to BEEC. The intervention focused on recognising and verbalising positive and negative emotions for the patients to become more confident with their own feelings.
(3) Emotional/sexual relationships. In this phase, it is best not to distinguish between affect and sexuality. This binomial should be used as a guide to become aware of the body, for example through masturbation and autoeroticism, as it allows individuals to embrace the pleasure and fantasies that originate from such actions. This phase is fundamental and must be accompanied by correct information because boys/girls can become more confident with regard to pleasure and feel the reactions of their own bodies, thus avoiding the cycle of thoughts related to their genital organs being different from other people. Re-establishing the significance of sexuality and emotions by observing the five dimensions of sexuality (Fig. 1), as described in the Del Re, Bazzo model [22,23,24] enabled work on the resources of the person.
(4) Building self-esteem. The focus on self-esteem was based on stimulating and fostering thoughts and reactions related to negative events. The authors worked together with each patient to find a new interpretation of said negative events. The aim of this phase was to give a meaning to the emotions that were felt and the resources used to face the events they emerged from.

Table 1 Questions and remarks that arose during the training.

Can you explain how I was born?
Will the catheter get in the way during sex?
I would like a longer penis
Who will accept me with such a small penis?
How can I get over the shame of having scars all over my body?
Can I give and receive pleasure?
Will a boy accept me with all of these problems?

Results

The SESAMO test was chosen because it can detect potential physical and psychological issues linked to the psychosexual-relationship sphere [18]. The SESAMO areas that presented a percentile rank that was greater than the 50th percentile were considered areas to be monitored, and those that were ≥70 were investigated in depth. Before the intervention, the areas above the 70th percentile could be identified as follows: related psycho-environmental, body-experience-related, areas of pleasure, medical and sexual history, motivation and conflict. The significant areas in the pre-intervention all fell below the 50th percentile in the postintervention period, both for male and female participants. More specifically, after the intervention, participants changed their attitude in several psychosexual areas, especially referring to psycho-environmental situations from the pre-intervention 84th median percentile rank for males (89th for females) to the postintervention 37th median percentile rank; in areas

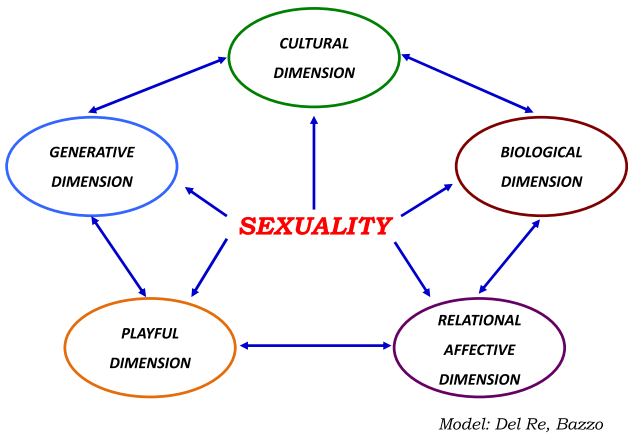

Figure 1 Del Re and Bazzo model of sexuality.

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Table 2 Areas, significant median percentile rank pre-intervention and postintervention.

Areas	Median percentile rank-pre pre-intervention		Median percentile rank-post postintervention	
	M	F	M	F
Related psycho-environmental situations	84*	89*	37	37
Body experience	98*	98*	46	38
Psychosexual identity	45	44	45	44
Desire	29	48	29	28
Areas of pleasure	90*	90*	37	37
Remote masturbation	66	52	49	33
Previous experience	62	54	48	45
Medical and sexual history	99*	98*	40	40
Motivation and conflict	95*	95*	31	31
Total	95*		37	

related to body experience, a drop was observed from the pre-intervention 98th median percentile rank to the postintervention 46th (38th for female) median percentile rank; in areas of pleasure, participants went from the pre-intervention 90th median percentile rank to the post-intervention 37th median percentile rank; in medical and sexual history, the drop was from 99th (98th for female) median percentile rank to 40th median percentile rank; and in motivation and conflict area, from 95th median percentile rank to 31st median percentile rank (Table 2).

Discussion

As shown in the literature, adolescents with BEEC suffer from significant physical problems that may interfere with their psychological development [16]. Body image, self-esteem, sexuality and sexual function are very important for adolescents, and this study provided a pioneering example of a program tailored to specific psychosexual topics concerning young people with BEEC.

The present study demonstrated, for the first time, that a targeted program may significantly improve the psychosexual condition of adolescents with BEEC. In particular, the study showed the need for adolescents to tackle topics of a psychological and sexual nature, as well as receive understandable answers with clear instructions they can be put into practice in their everyday lives. This could prevent patients from shutting themselves off, avoiding experiences with their peers, and being subject to the risk of developing further problems connected to their psychological and social discomfort. The interventions that were carried out had a specific and circumstantial focus on the life of adolescents. This enabled them to be provided with readily available tools to transform the knowledge they received throughout the program into actions and, therefore, improve the quality of their lives.

Admittedly, the study had several methodological limitations: (1) the research only involved 13 patients, so a greater number of participants would have certainly improved the level of representativity of the results, thus adding to their significance; (2) the SESAMO test was re-administered only shortly after the expert's advice had been given, and a longer interval between the end of the program and the follow-up test may have yielded more positive results, as the patients would have had more time to turn the acquired theoretical information into concrete actions; (3) the study had an exploratory nature and lacked counter-testing via a control group. In this regard, it is felt that it is important to underline that the study was designed using a test-retest methodology, without any control group, with the aim of controlling specific variables related to self-esteem, sexuality and emotions in adolescents affected by BEEC exclusively.

Despite these limitations, it is believed that this program represented a successful first attempt at contrasting the psychological problems of patients with BEEC. Using specific and targeted actions, it tried to put in place a series of preventative measures that may reduce the risk of social and psychosexual problems arising in relation to this medical condition.

Conclusions

This intervention methodology can be considered as a first attempt at improving support interventions for adolescents with BEEC, with the aim of enhancing their self esteem and contrasting forms of psychological difficulties in order to improve their quality of life.

Conflict of interest statement

Only University Institutional Resources were used to run this study, we have no conflict of interest to disclose.

The study was approved by the Independent Committee for Bioethics of the Institute for Maternal and Child Health — IRCCS 'Burlo Garofolo'.

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