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Evaluation of sexual function in females with exstrophy-epispadias-complex: A survey of the multicenter German CURE-Net

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Summary

Objective
Standardized knowledge about genital function in adult female individuals with exstrophy-epispadias complex (EEC) is scarce. The aim of this study was to investigate sexual function using the standardized Female Sexual Function Index (FSFI), and to assess the influence of bladder and vaginal reconstruction and the presence of incontinence on FSFI results.

Method
Sixty-one females (aged ≥18 years) recruited by the German multicenter network for congenital uro-rectal malformations (CURE-Net) were asked to complete the FSFI and a self-designed semi-structured questionnaire assessing comprehensive medical data, gynecological, and psychosocial items. Twenty-one eligible females (34%) returned both questionnaires (mean ± standard deviation [SD] age of 26 ± 5.1 years).

Results
In 43% of participants, a staged or single-staged approach had been used for reconstruction, and these had their bladder in use. A primary or secondary urinary diversion (UD) after cystectomy had been performed in 38% of participants. Of the participants, 57% lived in a committed partnership, and 62% had sexual intercourse on a regular basis, with a further 19% experiencing pain or discomfort thereby. Introitus plasty was done in 43%. Mean total FSFI for all participants was 21.3 (SD 1.9). Most domain scores of patients after introitus plasty were similar compared with those without an operative vaginal approach, except for satisfaction ($p = 0.057$) and pain ($p = 0.024$). Comparing incontinent with continent patients, significant differences were found for desire (mean 4.6 vs. 3.5, $p = 0.021$), lubrication (mean 3.1 vs. 4.2, $p = 0.049$), and satisfaction (mean 1.6 vs. 3.6, $p = 0.0065$). In contrast pain was not significant between groups.

Conclusions
Sexual activity rate in the present study was similar to that reported in the literature (81% vs. 89%), whereas dyspareunia rate was lower in our cohort (19% vs. 24%). The risk for sexual dysfunction seems to be lower in patients reconstructed with primary or secondary UD than patients with bladder in use. It is surprising that lubrification was better after UD than after bladder neck surgery. Incontinence and in some parts the history of an introitus plasty may play an additional role in development of sexual dysfunction in EEC. Although most of the female EEC patients lived in a committed partnership and had sexual intercourse, total FSFI values <26.55 clearly indicate a risk of sexual dysfunction. Although continence itself played a major role, females reconstructed with UD seem to have better sexual function. Further evaluation of sexual outcome and improvement of care for these patients is mandatory.

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Introduction

Today, long-term outcome issues such as sexual function represent the mainstay in adult patient care for female individuals with exstrophy-epispadias complex (EEC). There is no doubt, that sexual function in general is a complex and comprehensive issue affected by multiple, hardly measurable influencing factors. From the physical point of view, for years female genital reconstruction in EEC was judged to be more easy than male reconstruction; however, long-term physical outcome of female genital reconstruction was not reproducibly defined by objective or subjective criteria. In the literature, the main assessment criteria were satisfaction with genital appearance, sexual activity, own children, and relationships such as marriage. Additionally, quality-of-life-affecting factors such as urinary continence status, any restriction or discomfort in genital sensation, pain, genital prolapsed, and other unknown conditions may influence sexual activity in females with EEC. In recent years, a few well-designed single-center studies have described female sexual function in EEC with standardized instruments [1—3].

The aim of this study was to evaluate the sexual function in adult females with EEC using the German valuated Female Sexual Function Index (FSFI). As a secondary aim, the influence of continence status and kind of reconstruction on sexual function was investigated. For this analysis, patient data of the German network for congenital uro-rectal malformations (CURE-Net, www.cure-net.de) were used.

Materials and methods

Study population

CURE-Net is a multicenter population-based study from Germany, investigating environmental and genetic risk factors, clinical implications, psychosocial and psychosexual outcome for congenital uro-rectal malformations. In CURE-Net individuals with EEC have been recruited through participating departments of pediatric urology and pediatric surgery all over Germany and the German self-help organizations since 2009. The pseudonymized epidemiological and clinical data are stored in a centralized database. In a correspondent follow-up study over the period June—December 2014, 61 EEC female patients, aged at least 18 years in 2014, were recontacted by a postal invitation letter and asked to fill out two questionnaires to evaluate sexual function. Patients were excluded from the study in case of having been lost to follow-up ($N = 27$), having moved somewhere else ($N = 12$), or having returned only one of two questionnaires ($N = 1$). Written informed consent was obtained for each patient. This sub-study was approved by the ethics committee of the University of Ulm.

Data collection

A semi-structured questionnaire, containing 42 self-reporting items, was created by investigators to assess comprehensive medical data of participants such as phenotype, previous operative procedures, current continence status ("Are you incontinent [not to be able to control the discharge of urine]?", yes/no), voiding mode, gynecological history, prior pregnancies, child wish, specific sonographic results, and psychosocial items such as partnership-status (Appendix A). Additionally, the validated German version of the Female Sexual Function Index (FSFI) [4] was used, containing 19 self-reporting items assessing six domains of female sexual function (desire, arousal, lubrication, orgasm, satisfaction, and pain) over the previous 4 weeks (Appendix B).

Statistical analysis

Descriptive data of the study population and their sexual function are presented using absolute and relative frequencies. Calculation of the FSFI scores was made according to Rosen et al. [4] (Appendix C). An individual domain score of zero indicates no sexual activity during the past 4 weeks. The total FSFI score based on the sum of the six individual domain scores. The higher the total score (maximum 36.0), the better is the sexual function in female patients; a value $\leq$26.55 was classified as having a risk of sexual dysfunction [5]. In addition, subgroup analyses were performed to compare the results of the FSFI scores a) between patients with their bladder in use and patients with primary or secondary urinary diversion, b) between incontinent and continent patients, c) the overlap between these groups, d) between urethral voiding and catheterizing, e) between the presence of a cutaneous stoma and no urinary stoma, and f) between patients after introitus plasty and those without an operative vaginal approach. Comparison was done using the Student *t*-test. Statistical significance was defined as $p < 0.05$. Analyses were performed using the statistics software SAS, version 9.2 (SAS Institute Inc., Cary, NC, USA).

Results

Patients' characteristics

Twenty-one (34%) of 61 contacted females born between 1975 and 1996 participated in this sub-study (mean $\pm$ standard deviation [SD] age of 26 $\pm$ 5.1 years). Most had classical bladder exstrophy ($N = 11$, 52%), followed by cloacal exstrophy ($N = 4$, 19%), and epispadias ($N = 3$, 14%).

Operative procedures

Data on operative approaches to the bladder were missing in four patients (19%). Nine (43%) patients were reconstructed in either a staged ($N = 6$, 29%) or single-staged approach ($N = 3$, 14%) with their bladder in use (Table 1). Five of the functional reconstructed participants (56%) had an augmentation to optimize bladder storage function. Previous reconstructive operations to the external genitalia or the vagina were reported by 16 women (76%), nine of whom specified having had a previous introitus plasty (Table 1). Unfortunately, the time between any