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Male Sexuality, Fertility, and Urinary Continence in Bladder Exstrophy-Epispadias Complex

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ABSTRACT

Background: The bladder exstrophy-epispadias complex is a rare congenital malformation associated with severe dysfunction of the genital and urinary tracts and requiring a staged surgical reconstruction.

Aim: The primary aims of this study were to report the sexuality, infertility, and urinary incontinence outcomes in a cohort of men born with bladder exstrophy-epispadias complex. The secondary aim was to highlight some predictive factors of infertility in this population.

Methods: We conducted a descriptive, cross-sectional study of men diagnosed with classic presentations of bladder exstrophy or epispadias.

Outcomes: Patients were asked to complete 4 validated questionnaires: the International Index of Erectile Function (IIEF)-5, the Erection Hardness Score (EHS), the Self-Esteem and Relationship, and the International Consultation Incontinence modular Questionnaire-Short Form. Fertility potential was assessed with semen analysis and a non-validated questionnaire.

Results: 38 Patients 18−64 years old (M [mean] = 32.2) completed the questionnaires. The average IIEF-5 score was 18.1/25 (ranging from 3−25; SE = 7.62), with results indicating that 55% of the sample had normal erectile function. Results also showed higher scores for patients with normal spermatozoa concentration (M = 22.75, SE = 1.89, $P = .08$) than for those with oligospermia (M = 17.30, SE = 8.53, $P = .08$). Results on the IIEF-5 also indicated higher scores for patients who conceived children without assisted reproductive technologies (ART) (M = 22.83, SE = 2.317, $P = .02$) than for patients without children (M = 15.76, SE = 8.342, $P = .02$). The average EHS was 3.43/4 (ranging from 1−4, SE = 0.9). EHS was higher for patients who had reconstruction than for patients who had cystectomy (M = 3.88, SE = 1.07 and 2.78, SE = 1.09, $P = .02$). The average total Self-Esteem and Relationship score was 67.04/100 (ranging from 10.71−96.43, SE = 22.11). The average total International Consultation Incontinence modular Questionnaire-Short Form score was 4.97/21 (ranging from 0−18, SE = 5.44), higher score indicating more urinary incontinence. Among the patients surveyed, 31.6% were parents at the time of study and 50% of them benefited from ART. With regards to the 14 semen analyses performed, only 7.1% produced normal results and 44.7% indicated that ejaculation was weak and dribbling.

Clinical Translation: Erectile function appears to be decreased and psychological aspects of sexuality indicate low self-esteem about sexual relationship. Although ethical problems could not allow prospective spermograms, our cohort is large enough to provide significant data.

Conclusions: Early sperm storage for future ART, sexual medicine management, and complementary genital reconstruction in adulthood constitute potential treatment options for this population. **Reynaud N, Courtois F, Mouriquand P, et al. Male Sexuality, Fertility, and Urinary Continence in Bladder Exstrophy-Epispadias Complex. J Sex Med 2017;15:314−323.**

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INTRODUCTION

The bladder exstrophy-epispadias complex (BEEC) is a rare congenital malformation of the lower abdominal wall affecting 0.15−1 individuals in 200,000 births.[1,2] BEEC results from failure of the cavitation ("zip-down") process of the pelvic and sometimes the abdominal organs.[3] It may therefore affect the abdominal wall, the pelvic ring, the bladder and the urethra, the genitalia, and sometimes the bowels.

Its etiology remains unknown[1] and 3 clinical phenotypes[1,2] are commonly described according to the level of blockade of the zip-down mechanism: epispadias, classic bladder exstrophy, and cloacal exstrophy. Epispadias involves an open urethra sitting on the dorsum of the genital tubercle, associated with variable sphincter dysfunction.[1,2] Classic bladder exstrophy is characterized by an open bladder and urethra, and is associated with a gap between the 2 hemipubis, along with abdominal wall defect.[1,2] Cloacal exstrophy is the most severe case of BEEC and refers to exstrophied and sometimes duplicates pelvic organs, including the bowel tract, as well as other possible anomalies.[1,2]

Early surgical management protects the bladder mucosa from inflammatory or mechanical alterations and may help with bladder development required to achieve future continence.[1] Depending on the strategy considered in childhood,[1,2,4] several surgical techniques can be used. These are based on multiple reconstructions, performed in one (complete primary repair of bladder exstrophy) or multiple (classic stepwise approach[5]) stages, and are commonly associated with continent urinary diversion (Mitrofanoff) and bladder augmentation procedures.[1,2,4]

Additional treatments are often needed, such as bladder neck injection to reinforce the outlet resistance, bladder neck closure, or incontinent urinary diversion when all conservative procedures have failed.[2,3] Dissatisfaction with the appearance of both external genitalia and the abdominal wall (umbilicus) is common among adolescents requiring additional plastic procedure, which can be performed concomitantly with other genitourinary reconstructive surgeries.[6−8,17]

Fertility and sexuality are 2 domains often affected in men with BEEC[9−11] due to various possible causes, including recurrent genital or urinary tract infections or surgical injury.[12−15] Moreover, incompetent bladder neck and abnormal posterior urethra can cause ejaculatory dysfunction,[15] while shortness of the genital tubercle can cause sexual dissatisfaction.[16,17] Patients may also experience difficulty with intromission due to a short and broad penis, or the existence of a penile dorsal curvature.[16−18] Finally, self-esteem can be impaired because of anxiety about genital appearance and sexual activity in general.[17]

This study aims at describing sexuality quality, fertility, and urinary status in a cohort of men born with BEEC. As a secondary objective, the study also aims at highlighting some predictive factors of infertility and comparing different types of surgery and related phenotypes.

METHODS

An analytical cross-sectional study was conducted at Lyon University Hospitals (Hospices Civils de Lyon) from June 2016−September 2016. The target population was men born with classic bladder exstrophy or epispadias. Exclusion criteria included children and adults under guardianship. Eligible participants were identified through the hospital database (Programme de Médicalisation des Systèmes d'information). They were all being followed up within 3 different departments at the same institution (adult urology, pediatric urology, neuroperineal and sexology) between January 2009−November 2015.

Eligible participants were instructed to complete 4 validated questionnaires about sexuality: the International Index of Erectile Function (IIEF)-5,[19] the Erection Hardness Score (EHS),[20] the Self-Esteem and Relationship (SEAR),[21] and the International Consultation Incontinence modular Questionnaire-Short Form (ICIQ-SF) assessing urinary status.[22] Fertility was assessed with spermograms and a non-validated questionnaire.

Sexuality

Erectile function was assessed using the IIEF-5, which is a 5-item self-report measure. Total scores range from 0−25 and reflect the presence and severity of erectile dysfunction (ED). According to this score, ED is classified as absent (21−25), mild (16−20), moderate (11−15), severe (5−10), or uninterpretable (1−5).[19]

Erection rigidity was assessed using the EHS. The EHS is a single-item self-report measure assessing erection hardness on a 0−4 scale. A score of 0 indicates the absence of any rigidity; a score of 1 is indicative of some tumescence but no rigidity; a score of 2 characterizes a rigid penis, yet not rigid enough for penetration; a score of 3 indicates a sufficient amount of penile tumescence for penetration without full rigidity; and a score of 4 reflects maximum hardness and rigidity.[20,23]

Sexual outcomes were assessed using the SEAR questionnaire, which is an ED-specific validated 14-item self-report measure. The SEAR questionnaire yields 5 different scores relating to psychosocial variables: a total score, a sexual relationship satisfaction domain score (items 1−8), a confidence domain score (items 9−14), including a self-esteem (items 9−12), and overall